

Children with spina bifida and their mothers in Palestine

Lived experiences, self-perceived health and sense of coherence

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Thesis Title

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For the soul of my mother and father

For my husband and my beloved children

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ABSTRACT

The overall aim in this thesis was to describe the lived experiences of children with spina bifida (SB) and their mothers while living under specific social and cultural conditions in West Bank, Palestine. A further aim was to gain knowledge about the impact of SB on self-perceived health and sense of coherence (SOC) among a group of children and adolescents with SB in comparison to their healthy peers.

Study I and II involved narrative interviews with 20 mothers and 10 children, respectively, which were analysed with the phenomenological-hermeneutical method. Study III and IV involved self-reported questionnaires. The Pediatric Quality of Life Inventory (PedsQL™ 4.0) was used in Study III to assess self-perceived health in 50 children with SB in comparison with 150 healthy children. In Study IV, the PedsQL™ 4.0 and SOC scales were used to assess self-perceived health and SOC in 50 adolescents with SB in comparison with 150 healthy adolescents. The association between self-perceived health, SOC and levels of impairment in children and adolescents with SB was also investigated. The findings in this thesis show that children with SB, their mothers and other family members were struggling with negative sociocultural beliefs about the disability. Stigmatisation, political conflict and consequent poor health care services were shaping the daily life experiences of the children and their families. The lack of knowledge about health care needs of the child with SB had a true effect upon the mothers' well-being, and increased their burden. Children with SB were struggling with social exclusion and seemed to possess a negative self-concept. Children and adolescents with SB perceived their health to be worse than the health of their peers. SOC was lower in adolescents with SB than it was in their healthy peers. Levels of impairment in mobility and hydrocephaly associated negatively with self-perceived health in children and adolescents with SB, while bladder dysfunction was negatively associated with SOC only among the adolescents.

In conclusion, stakeholders, policy makers, health care providers and educators in Palestine must be aware of the extent of the burden and suffering experienced by children with SB and their families. Health promotion and social support for this group of children and their families can be achieved through the development and implementation of effective policies and strategies, such as a family-centred care (FCC) approach, within health care systems and rehabilitative practices in the Palestinian context.

Keywords: Childhood, Disability, Experience, Family, Mother, Palestine, Perceived health, Phenomenological-hermeneutical, Spina bifida, Stigma, Sense of coherence

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SAMMANFATTNING PÅ SVENSKA

Det övergripande syftet med denna avhandling var att beskriva de levda erfarenheterna hos barn med spina bifida (SB) och deras mödrar under de specifika sociala och kulturella förhållandena i Västbanken, Palestina. Ett ytterligare syfte var att få kunskap om SB:s inverkan på självskattad hälsa och känsla av sammanhang (SOC) hos barn och ungdomar med SB jämfört med deras friska kamrater.

Studie I och II involverade narrativa intervjuer med 20 mödrar respektive 10 barn, vilka analyserades med fenomenologisk-hermeneutisk metod. Studie III och IV involverade självrapporterad data i form av enkäter. Pediatric Quality of Life Inventory (PedsQL 4.0) användes i studie III för att bedöma självskattad hälsa hos 50 barn med SB jämfört med 150 friska barn. I studie IV användes PedsQL 4.0 och SOC-13 skalan, för att mäta självskattad hälsa och SOC hos 50 ungdomar med SB jämfört med 150 friska jämnåriga. Associationen mellan självskattad hälsa, SOC och funktionsnedsättning hos barn och ungdomar med SB undersöktes också. Resultaten i denna avhandling visar att barn med SB, deras mödrar och övriga familjemedlemmar kämpade med negativa sociokulturella uppfattningar om funktionshinder.

Stigmatisering, den pågående politiska konflikten och därmed bristfällig hälso- och sjukvård, påverkade barnens och deras familjers erfarenheter. Bristen på kunskap om barnens behov av vård och rehabilitering påverkade mödrarnas välbefinnande och ökade deras börda. Barn med SB kämpade med social utestängning och tycktes ha en negativ självbild. Barn och ungdomar med SB upplevde att deras hälsa var sämre än deras jämnårigas hälsa. SOC var lägre hos ungdomar med SB än den var hos deras friska kamrater. Nivån av nedsatt mobilitet och hydrocefali associerade negativt med självskattad hälsa hos barn och ungdomar med SB, medan blåsdysfunktion var negativt korrelerad med SOC endast bland ungdomarna.

Sammanfattningsvis måste intressenter, beslutsfattare, vårdgivare och lärare i Palestina vara medvetna om omfattningen av den börda och det lidande som barn med SB och deras familjer upplever. Hälsofrämjande åtgärder och socialt stöd till denna grupp av barn och deras familjer kan uppnås genom utveckling och genomförande av effektiva strategier, såsom familjecentrerad vård, inom hälso- och sjukvård och rehabilitering i Palestina.

ملخص باللغة العربية

الهدف العام من هذه الرسالة هو وصف تجارب الحياة اليومية لدى الأطفال الذين يعيشون مع مرض الصلب المشقوق وعائلاتهم وتأثير ذلك على النظرة الشخصية لصحتهم ومدى التماسك والتحكم والثقة بالنفس لديهم. لقد تم استخدام نهج متعدد الأساليب لجمع وتحليل البيانات.

في الدراستين I و II تم عقد مقابلات سرديّة مع 20 أم و 10 أطفال على التوالي وتم تحليلهم بطريقة تحليل البحث النوعي phenomenological Hermeneutical. لقد تم استخدام استبيان PedsQL في الدراسة III لقياس الصحة الجسدية الذاتية ل 50 طفل مصابين بمرض الصلب المشقوق ومقارنتها مع 150 طفل أصحاء. في الدراسة IV تم استخدام نفس الاستبيان بالإضافة الى استبيان ال SOC لقياس الصحة الجسدية والقدرة على التماسك والتحكم والثقة بالنفس ل 50 مراهق مصابين بمرض الصلب المشقوق ومقارنتها مع 150 من المراهقين الأصحاء. بالإضافة الى دراسة العلاقة بين مستوى الإعاقة لدى الاطفال ونظرتهم للصحة الجسدية والثقة والتماسك.

اشارت النتائج إلى أن وصمة العار والفقر وضعف الخدمات الصحية والحماية الاجتماعية في ظل الظروف السياسية السائدة تسيطر على الحياة اليومية للأطفال وأمهاتهم. وان نقص معرفة الأمهات باحتياجات الطفل المصاب بمرض الصلب المشقوق يؤثر على تقديم الرعاية الصحية اللازمة للطفل ويؤثر على صحة الأم النفسية ويزيد من أعبائها. اما بالنسبة للأطفال المصابون بمرض الصلب المشقوق فقد كانوا يعيشون في صراع نفسي ويعانون من الإقصاء الاجتماعي. كما اشارت الدراسة ان لديهم تصور للصحة الجسدية والتماسك والثقة بنسبة اقل بكثير من الأطفال الاصحاء. توصلت الدراسة الى وجود علاقة سلبية بين مستوى الإعاقة ونظرة الأطفال للصحة الجسدية والتماسك. في الختام تفيد هذه الدراسة في زيادة الوعي حول المشاكل الاجتماعية والنفسية التي يعاني منها الأطفال لمصابون بمرض الصلب المشقوق وأسرههم. ويطلب من صانعي السياسات، ومقدمي الرعاية الصحية والمعلمين في فلسطين أن يكونوا على وعي تام بخبرة هؤلاء الأطفال وتجاربهم كما ينصح بتحقيق

الدعم الاجتماعي وتعزيز الصحة لهذه المجموعة من الأطفال وأسرهم من خلال وضع استراتيجيات
فعالة ومتطورة مثل ممارسات الرعاية بالأسرة لإعادة التأهيل.

LIST OF PAPERS

This thesis is based on the following studies, referred to in the text by their Roman numerals.

- I. Nahal, M. S. H., Wigert, H., Imam, A., & Axelsson, Å. B. (2017). From feeling broken to looking beyond broken: Palestinian mothers' experiences of having a child with spina bifida. *Journal of Family Nursing*, 23(2), 226–251
- II. Nahal, M. S. H., Axelsson, A. B., Imam, A., & Wigert, H. (2019). Palestinian children's narratives about living with spina bifida: Stigma, vulnerability, and social exclusion. *Child: Care, Health and Development*, 45(1), 54.
- III. Nahal, M. S. H., Wigert, H., Imam, A., & Axelsson, Å. B. (2019). Self-perceived health among children with spina bifida in the West Bank, Palestine. Manuscript.
- IV. Nahal, M. S. H., Wigert, H., Imam, A., & Axelsson, Å. B. (2019). Assessment of health status in adolescents with spina bifida in the West Bank: Sense of coherence and self-perceived health. Manuscript.

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ABBREVIATIONS

CBR	Community Based Rehabilitation
CIC	Clean Intermittent Catheterization
CWDs	Children with Disabilities
FCC	Family Centred Care
GCS	Generic Core Scale
HRQoL	Health Related Quality of Life
NGOs	Non-Governmental Organisations
NTDs	Neural Tube Defects
PA	Palestinian Authority
PCBS	Palestinian Central Bureau of Statistics
PedsQL	Paediatric Quality of Life Inventory
PMOH	Palestinian Ministry of Health
PRO	Patient Reported Outcomes
QoL	Quality of Life
SB	Spina Bifida
SOC	Sense of Coherence
UN	United Nations
UNRWA	United Nations Relief and Works Agency
WHO	World Health Organisation

INTRODUCTION

Research on the experiences of children with chronic illness and disabilities has increased in recent decades, and it has been documented that conditions such as spina bifida (SB) have a lifelong impact on the health and quality of life (QoL) of children (Bakaniene, Prasauskiene, & Vaiciene Magistris, 2016). Despite advances in treatment, children often experience motor impairment, paralysis, hydrocephaly, chronic bowel and bladder incontinence, seizures, orthopaedic deformities, and cognitive and vision deficits throughout their lifetime (Psihogios, Kolbuck, & Holmbeck, 2015). These consequences can have a significant and cumulative impact on psychosocial health and the functioning of families (Copp et al., 2015).

As a paediatric nurse, I have worked with children of all ages. I have provided information, care, support and guidance to children and their mothers, including children with SB who were admitted to the hospital for evaluation and management. My experiences as a hospital paediatric nurse have made me reflect on the complexities surrounding children with SB and their mothers, who might have been at risk. I have paid attention to mothers who ask many questions and who lack knowledge about their child's condition. My work has indisputably brought me closer to them, and I have learned much about the difficulties they face. Yet, what struck me most about the care provided for these children is that we often focus on basic hospital care and medical aspects, with less focus on long-term follow-up care, daily life functioning, psychosocial concerns and well-being. In the absence of the recognition of these problems within health care services in Palestine, I felt that it was my humanitarian duty to highlight these problems.

The present study takes its starting point within health care sciences, via a holistic approach, by means of describing the lived experiences of children with SB and their mothers. Knowledge about how life is experienced from an individual's perspective is needed to plan for lifetime management and to customise care interventions throughout the whole lifespan (Ekman et al., 2011). This thesis offers an opportunity to explore how Palestinian children with SB and their mothers experience and handle their daily lives. Further, the thesis investigates the impact of SB on children's and adolescents' health and well-being while living under the specific sociocultural and political conditions in Palestine.

1 BACKGROUND

1.1 SPINA BIFIDA

Spina bifida (SB), also called myelomeningocele, is one of the major neural tube defects (NTDs) and has been described as the most complex birth defect that can be survived beyond childhood (Liptak, Garver, & Dosa, 2013). SB involves a split (bifid) in the spinal column that occurs in the fourth week of pregnancy and results in the protrusion of the meningeal sac with portions of the spinal cord or spinal nerves (Copp et al., 2015). The sac can occur anywhere along the spinal column but occurs more frequently in the lumbar region. The condition causes complex neurocognitive impairment as well as functional problems in affected individuals that vary in severity depending on the magnitude and location of the lesion on the spine. The condition can thus affect the psychological and social profile of sufferers (Hockenberry & Wilson, 2018).

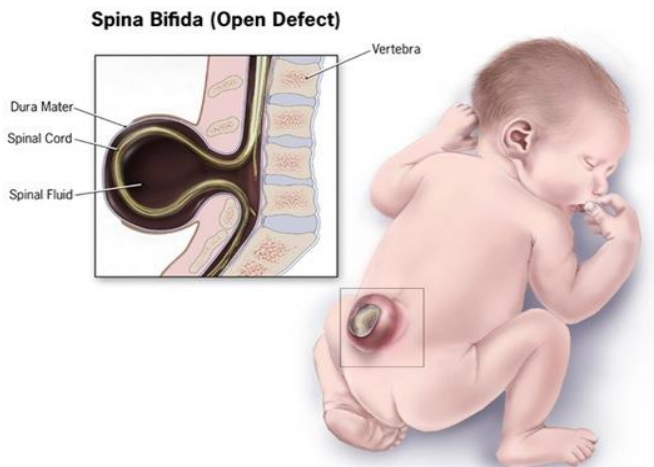


Figure 1. SB open defect (Retrieved from [www. healthjade.com/spina-bifida](http://www.healthjade.com/spina-bifida)).

The aetiology of NTDs is still debated. The most important and well-known risk factor associated with SB is folic acid deficiency in mothers (Wilson et al., 2015). Other suggested factors include geographic variations, consanguineous marriage, genetics, nutritional, environmental and teratogenic factors, gestational diabetes, maternal age, obesity and anticonvulsant drugs (Copp et al., 2015).

Over the last two decades, the incidence of SB has dropped significantly in many countries, probably due to improved maternal diet, pre-conception folic acid supplementation and improved antenatal screening (Youngblood et al., 2013). However, elective termination of pregnancy makes it difficult to ascertain the actual incidence of SB (Jenkinson et al., 2011). The worldwide prevalence of SB ranges from 10–100 per 10000 live births, and more than 300,000 cases are estimated to occur annually (Au, Ashley Koch, & Northrup, 2010). Many of these cases occur in less-developed, low-income countries in Latin America, Africa, Asia and the Middle East, including Palestine (Fonseca, Raskin, & Zugaib, 2013).

1.1.1 GROWING UP WITH SPINA BIFIDA

Babies born with SB typically undergo neurosurgery to close the defect and prevent further infection and damage (Trudell & Odibo, 2014). Advancements in surgical repair have decreased the mortality and morbidity rate of affected individuals. Today, patients with SB have an almost normal life expectancy (Vande Velde et al., 2016).

These patients present a wide spectrum of motor and sensory impairments that vary in nature and severity. The primary functional deficits are lower limb paralysis, sensory loss, bladder and bowel dysfunction, hydrocephaly and some cognitive deficits (Copp et al., 2015). Patients with SB require ongoing neurosurgical and medical attention (Greenley, 2010). Thus, despite advancements in surgical repairs, children grow up with serious physical and neurocognitive conditions (Dicianno et al., 2008).

Although children with SB often display average intellectual functioning, they are at risk of learning problems (Vinck et al., 2010). A previous literature review of the cognitive phenotype showed poor executive skills in children with SB as well as difficulties in perception, language, literacy, attention, memory and numeration (Dennis & Barnes, 2010). They often experience delayed and limited self-care, participation in daily life and autonomy (Peny-Dahlstrand, Krumlinde-Sundholm, & Gosman-Hedström, 2012).

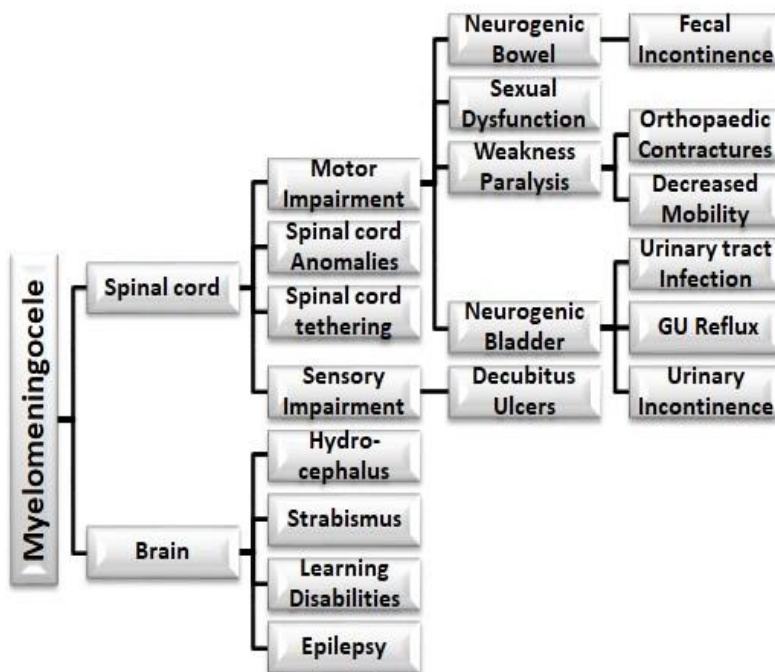


Figure 2. A flow chart about the problems associated with the SB defect (myelomeningocele).

1.1.2 IMPACT OF SPINA BIFIDA ON CAREGIVERS

The increasing survival rate of children with SB poses a global challenge to health care systems to meet the physical and psychosocial needs of these children and their families (Sawyer & Macnee, 2010). The complex consequences of SB have implications not only for the affected children but also for their parents and other family members (Holmbeck & Devine, 2010) severely impacting their health and social lives; this is especially true for mothers, who take an active role in managing their children's care (Ong, Norshireen, & Chandran, 2011). While mothers can develop special competences in managing their children's care, their caring role is undervalued (Ryan, 2015). Previous studies have explored the mothers' feelings, as mothers often experience more intense feelings and psychosocial stress than do other

family members (Ong et al., 2011). In Coughlin and Sethares (2017) study, mothers of children with chronic illness and/or disabilities exhibited significant feelings of fear, guilt, self-blaming and sorrow. The mothers' need for continuous support was emphasised because of the serious demands placed upon them and other family members (Bannink, Idro, & van Hove, 2016).

It is known that the stigma of disability, particularly in developing countries, can affect the psychosocial health and well-being of children with SB and their families (Lang, Kett, Groce, & Trani, 2011; Lindsay, 2014), placing them at risk of social exclusion (Rofail et al., 2012). Accordingly, evaluating the psychosocial aspects of children and their families has recently been emphasised (WHO & Unicef, 2012) to promote their rights, health and functioning (UNCRPD, 2006).

1.1.3 HEALTH CARE MEASURES OF CHILDREN WITH SB

Management of children with SB requires a multidisciplinary approach involving a clinical nurse specialist or nurse practitioner; paediatric specialists in neurosurgery, orthopaedics, urology, developmental paediatrics and physical medicine; physical therapists; orthotists, psychologists and social workers; and health education professionals (Burke & Liptak, 2011). Achieving optimal functioning in children with SB involves the coordination of care and a flexible and dynamic partnership between clinicians, parents and children (Hockenberry & Wilson, 2018).

Nurses play an important role in caring for children with SB and in assessing parents' ability and willingness to nurture and care for these children. The nurse coordinator participates in a number of intervention strategies to prevent long-term disability, such as performing neurological assessments and dealing with orthopedic, urinary and bowel complications (Dunleavy, 2007). Bladder function can be improved by the early introduction of clean intermittent catheterization (CIC) and by teaching families and children (when older) about the technique (Hockenberry & Wilson, 2018). Bowel management requires a regular toilet-training programme early in development to help the child develop and maintain regular bowel movements. Hydrocephaly is controlled by assessing and detecting early signs of shunt failure and by teaching the family about these signs (Burke & Liptak, 2011). Early orthopedic assessment and physiotherapy are also important to maintain the strength and flexibility of muscles and to prevent deformities.

1.2 DISABILITY

Disability can be viewed as a complex phenomenon, not just a health problem. It can be defined as ‘individual loss or abnormality of a body part or functioning, i.e. physical or cognitive functioning’ (WHO, 2011) which has a substantial and long-term adverse effect on the individual’s ability to carry out normal day-to-day activities (Richardson, 2010). Disability is also defined as impairment (a problem in body function), activity limitations (difficulty to perform action), and participation restrictions (difficulty to be involved in life situations) (Leonardi, Bickenbach, Ustun, Kostanjsek, & Chatterji, 2006).

Disability can also be explained from the perspective of the socio-ecological model, which distinguishes between the impairments people have and the harassment they experience as a result (Shakespeare & Watson, 2001). This indicates that impairment is becoming a disability through the experience of ‘structural oppression; including stereotypes, socio cultural attitudes, bureaucratic hierarchies, political and economic status, and all that is relating to how society is structured and organized’ (Thomas, 2010, pp 42-43).

Worldwide, people with disabilities face challenges related to poor health outcomes, as well as economic and social disadvantages. This is partly because they experience barriers in accessing health, education, employment and transport services. These difficulties are expected to be worse in developing and low-income countries, such as Palestine (Jarar, 2009). The children’s right to be free from discrimination and social isolation has not yet been achieved, despite the United Nation’s (UN) convention on the rights of children with disabilities in Palestine (UNICEF, 2016).

1.3 THE PALESTINIAN CONTEXT

Children with chronic illness and disabilities experience different situations across societies due to variations in sociocultural issues, health care provisions, rehabilitation systems, religions and family dynamics (WHO, 2011). To understand the experiences of children with SB and their families in West Bank, Palestine, the context in which they live should be explored.

Palestine has been under Israeli control since 1948. Today, Palestine can be described as a state geopolitically segregated into two zones: the West Bank (including East Jerusalem) and the Gaza Strip. Palestine’s current estimated population is 4.74 million, 2.89 million of whom live in the West Bank, while 1.85 million live in Gaza. The majority of the Palestinian population are young:

children aged 0–14 comprise approximately 39% of the population (Palestinian Central Bureau of Statistics, 2016).

1.3.1 POLITICAL ISSUES IN PALESTINE

The protracted political conflict (Israeli–Palestinian conflict) in the state of Palestine is considered a serious threat that affects several aspects of life of the Palestinian population (Giacaman et al., 2009). Yet, despite the establishment of the Palestinian Authority (PA), as outlined in the 1994 Oslo Peace Accords, the full governance and control of the land, borders, basic resources and water, as well as people’s movement and many other social determinants of health, are still lacking (Giacaman et al., 2009).

The PA faces a variety of significant challenges due to the ongoing political conflict and economic instabilities, which have caused difficulties in building the basic infrastructure of the state. Severe political pressure, siege and the construction of the annexation wall by the Israelis have imposed serious constraints on the Palestinian people, consequently impacting their health, education and socioeconomic status as well as depriving them of their basic human rights (Isaac & Hilal, 2011).

1.3.2 THE FAMILY WITHIN THE PALESTINIAN CONTEXT

The family is the ‘basic social unit in traditional and contemporary Arab society’ (Barakat, 1993, p. 23); in Palestine, it is the core social institution. Arab culture greatly influences Arab families’ lives, as does the Islamic faith. Cultural beliefs of loyalty, cohesiveness and solidarity to the family in Arab societies stem from Islamic religious beliefs and are considered a source of strength, protection and security for all family members, including mothers and children (Hanan et al., 2009). Whilst it is true that the contemporary Arab family unit has been influenced by current political and economic developments and is progressively moving to a nuclear form – at least in urban areas – the links between extended families remain strong and important (MacGregor & Godfrey, 2011).

Traditional families in Arab societies are highly patriarchal, characterised by a sexual division of work and by men’s power over women. The strongly gendered social roles in Palestine increase the burdens placed on women in many aspects of life. Mothers in Arab society are typically the primary caregivers of children, while fathers tend to be less involved with childcare responsibilities (Hasnain, Shaikh, & Shanawani, 2008). Further, mothers tend

to tolerate and accept social blame for and negative social attitudes about giving birth to children with disabilities (Crabtree, 2007).

That said, Arab families are not, of course, all identical. Recent changes in family structure have contributed to the democratisation of husband–wife and father–children relationships. According to Barakat (1993, p23), women’s education and employment contributes to the transition towards democratic relations inside the family. Increasingly, the father’s role is changing to one that is more involved in the family and shares authority and responsibility with other family members. Yet, the family remains patriarchal and hierarchical in structure.

1.3.3 ISLAM AND SOCIOCULTURAL ATTITUDES

Islam is a crucial factor in all aspects of Palestinian life, as 94% of the population is Muslim (Giacaman et al., 2009). In Palestine, as well as in most Arab Muslim countries, religious, cultural and political forces are often intimately entwined in the formulation of people’s beliefs.

As Hasnain et al. (2008) described, the main aspects of Islamic principles of disability are justice, equality and non-discrimination. It is indicated in the Quran that a Muslim’s worth is not based on physical or material characteristics but rather on piety, faith, prayer and adherence to Islam’s obligations to the best of one’s ability. Islamic education encourages Muslims to care for people with disabilities, to handle their affairs, to be kind to them and to treat them with dignity. Therefore, Muslim families are required to care for their children with chronic illness or disabilities, to have patience and to endure pain as a sign of strong faith, asking for God’s mercy and forgiveness.

Although Islamic principles urge Muslims to care for people with disabilities, negative traditional attitudes can influence people’s beliefs and behaviours. Some traditional attitudes view disability as being the result of sins and mistakes committed by a family member in a previous life or as a punishment from God (Hasnain et al., 2008). Therefore, some parents prefer to hide their disabled children from society or avoid seeking care and governmental services due to fear of humiliation, stigma and disgrace, leading to unnecessary suffering in children and their families (Burton, Sayrafi, & Srour, 2013). Few efforts have been made by Muslim societies to improve this situation on a national level, and most approaches for challenging the stigma of disability were originally developed and adapted for Western developed communities (WHO, 2011). Khamis (2008) argued that there has been a gradual reduction in stereotyped perspectives on children with disabilities (CWDs) in Palestine, likely due to the increased number of CWDs as a result of the ongoing conflict

with Israel. Caring for these children has become a national responsibility, as they have been depicted as strugglers and have attained a martyrdom status. However, Burton et al. (2013) emphasised that reducing social stigma will require much more effort by the PA, because martyrdom disabilities will not put an end to the social discrimination of people with other disabilities.

1.3.4 DISABILITY IN THE PALESTINIAN CONTEXT

Current statistics about people with disabilities in the West Bank indicate that seven percent of the population, or 202,000 individuals, are living with some kind of disability. The prevalence of disabilities among children aged 0–17 years is 1.6%; this means that 47,000 children currently live with severe forms of disability in the West Bank (Palestinian Central Bureau of Statistics & Ministry of Social Affairs, 2011). The PA is working to improve the lives of people with disabilities, particularly children, by establishing a special department to follow and resolve relevant issues under the umbrella of the Ministry of Social Affairs and by the ratification of an international convention of rights of persons with disabilities on April 2, 2014. Yet, practical evidence reveals major implementation setbacks (Jarar, 2009).

UNICEF (2016) studies in Palestine have reported that meeting the needs of people with disabilities is still lacking, with the right of full access to health, schools, social inclusion and other aspects of society not yet being achieved. The latest census of disabilities showed that 38% of CWDs have never enrolled in school, while 34% have dropped out of school; further, 87% of those of working age are unemployed. It was also reported that 74% of disabled persons with mobility issues suffer from lack of access to public services (Palestinian Central Bureau of Statistics & Ministry of Social Affairs, 2011).

CWDs worldwide face challenges of discrimination and violations of their right to healthcare, education and even survival, thereby undermining their social inclusion (WHO, 2011). Palestinian CWDs are facing a particularly dire situation, as they live in an area prone to violence and political conflict. According to the UNICEF (2016) report, the protracted political conflict and cultural stigma in Palestine were identified as major obstacles to promoting the rights of CWDs. Further, Burton et al. (2013) argued that protecting children from violence and changing social attitudes towards disability are urgently needed in Palestine.

1.3.5 HEALTH CARE SYSTEM IN PALESTINE

There are four major providers of health services in Palestine: (1) the Ministry of Health (PMOH) provides primary, secondary and tertiary health services

and purchases advanced health services from domestic and foreign providers ; (2) Palestinian non-governmental organisations (NGOs) provide primary, secondary and some tertiary services; (3) the United Nations Relief and Works Agency (UNRWA) provides primary care services for refugees and can purchase services for hardship cases; and (4) the private sector, which provides the three levels of care through a variety of specialised hospitals and investigation centres (Mataria et al., 2009). However, effective communication and coordination between these different sectors are lacking, as are the services provided for children with mental health issues or disabilities (Waterston & Nasser, 2017).

1.3.6 REHABILITATION SERVICES

In 1991, the disability sector in Palestine began transitioning from traditional ways of caring for CWDs (i.e., charitable, medical and religious obligations) to community-based rehabilitation (CBR) services (Giacaman, 2001). The aim of CBR is to empower people with disabilities to access health, education, employment and social services with equal rights to social opportunities (UNICEF, 2016). Further, CBR seeks to change cultural attitudes, ensure social integration, improve capabilities and promote the independence of CWDs (Burton et al., 2013; Eide, 2006). The main CBR services include home visits, exercise training in the home environment, and teaching and supporting CWDs and their families to help them take a leading role in the rehabilitation process. CBR providers take responsibility for communication and coordination with the local government and private rehabilitation centres to arrange for children's admission and follow-up care when needed (Nilsson & Qutteina, 2005).

However, CBR strategies face challenges that are mainly linked to political conflict and its consequences; these challenges prevent the PA from extending CBR services to broader communities. Consequently, the services are limited to a few urban areas, which means they are not available for all families. Further, CBR programmes give priority to conflict-related physical disabilities, such as spinal cord injuries, which in turn decreases opportunities for children with other types of disabilities to benefit from these services (UNICEF, 2016).

The accessibility of CBR services is another challenge. Although all CWDs are given free health insurance for CBR services, access to health care is still lacking. Recent studies in Palestine have shown that 50% of families with CWDs lack knowledge about disability-related forums and opportunities for their children to benefit from CBR services (UNICEF, 2016). While the CBR

programme covers 45% of all local communities in Palestine, the total number of CWDs served is limited. The current statistical evaluation indicates that only 1,412 people are enrolled in the programme's services. It was suggested by the UNICEF (2016) report that successful implementation of the CBR programme requires unified effort by CWDs, their families, communities, governmental and non-governmental health, education and social services.

1.3.7 PERSONAL COMMUNICATION WITH NURSES IN THE REHABILITATION CENTRE

Given the importance of nurses' participation in decision-making and their role in improving health care services (Gazarian, Henneman, & Chandler, 2010), I found it valuable to communicate with some nurses in one of the major rehabilitation centres responsible for providing services to patients with SB. The main aim of the discussion was to gain knowledge about the provision of care and the nurses' role in caring for children with SB in Palestine. Knowledge gained from these personal communications is exemplified in the following short summary:

‘Currently there is no specific rehabilitation centre for children with SB in Palestine, although we provide them with the needed services as much as possible when they are admitted to our centre. Children born with SB will be transferred to our centre for registry. The family will meet the specialist, who will give them explanations about the child's condition, discuss with them the future investigations, therapies and surgeries for the child. In the previous years and before the first intifada (1987), we had a structured rehabilitation programme with well-trained staff to address the specific needs of the patients with SB and enhance their daily lives. The programme was donated from the Swedish Diakonia to cover nearly all children with SB in the West Bank. However, this programme was not sustained long due to the increasing number of spinal cord injuries in the intifada, which has shifted our efforts to promoting the lives of injured youth who are increasingly admitted to our centre. Then, the donation was stopped, and the PA could not provide the centre with the needed facilities as before’.

‘We do know that SB is a life-long process, and its burdens can never be carried by the families nor by the present centres. Caring for these children requires the collaboration of all parties concerned in the community to meet the huge needs and costs

of care for these children. Today we have few beds for children with SB who are transferred here from the CBR services for learning about CIC or bowel training when they reach the age of seven years. We also receive children who need mobility rehabilitation, physiotherapy and occupational therapy, as well as patients with serious complications’.

These nurses showed concern about the current services provided for children with SB and their families in Palestine. They also emphasised the importance of collaboration with the CBR staff to exchange experiences and obtain knowledge about CBR services in order to improve the children’s conditions and maintain their health.

1.4 CENTRAL CONCEPTS

This thesis elucidates three overarching constructs. The first is the core block, which concerns lifeworld experiences. The second important construct is the theory of family-centred care (FCC), which may promote the caring paradigm among children with SB and their families. The third relevant construct in this study assessed how children with SB perceive their health by incorporating self-perceived health and sense of coherence (SOC).

Stigmatisation is a key component in this thesis insofar as it affects the lifeworld experiences of children with disabilities and their families. The threat of potential stigmatisation can complicate the promotion of health and well-being for children with SB and their families. In the following section, lifeworld experiences, family-centred care, self-perceived health, SOC and stigmatisation will be explored both in general and in relation to children with SB and their families.

1.4.1 LIFEWORLD EXPERIENCES

Edmund Husserl (1859–1938), philosopher and founder of phenomenology, introduced the concepts of subjectivity and ‘lifeworlds’, with the latter referring to meaning structures in which all individuals live unreflectively in a common world (Todres, Galvin, & Dahlberg, 2007). This means that we experience the everyday world through a ‘natural attitude’, an attitude in which we judge – and have already made judgements – about the existence of phenomena (Lindseth & Norberg, 2004). Husserl asserted that a human being is an indivisible whole, i.e., there is no subject–object split, but there is a subject–object correlation (Behnke, 2011).

Merleau-Ponty's argument, based in an ontological understanding of the lifeworld, indicated that human beings are in the world as 'flesh', and that a body is a prerequisite for being in the world (Merleau-Ponty, 2004, p 95). The phenomenological mission of Husserl was further expanded by Heidegger (1889–1976), who moved the focus from consciousness to existential and interpretive dimensions to reveal the hidden meaning of phenomena and explain the experience of 'being in the world' (Finlay, 2012).

Husserl's phenomenological understanding of 'lifeworld' was articulated in five elements: embodiment, temporality, spatiality, intersubjectivity and mood (Hemingway, 2011). Embodiment refers to the assumption of a lived body – the way a body is experienced. Further, individuals 'have not' merely a body but also 'are' their bodies. Bodies are main anchors and homes in life, i.e., they give us our direction and access to life. Extending this assumption, any alteration related to the body affects both one's lifeworld and opportunities for realising life. Temporality refers to how time is experienced rather than to objective time, while spatiality concerns how places and spaces are experienced. Intersubjectivity signifies relatedness to other people and how lived relations constitute experiences of the lifeworld, as a man is not an isolated island, but is instead constantly in communion with other humans. Mood refers to how the world is received and perceived from an individual's perspective. These elements are closely intertwined, which means that the concept of lifeworld is complex and dynamic, i.e., its horizons constantly broaden as we accumulate new experiences, in both health and illness, as well as in encounters with other individuals and with situations or challenges. A child with SB or another disability lives in the world with a disrupted body, which in turn affects his or her lifeworld and all its elements.

1.4.2 FAMILY-CENTRED CARE

The philosophy of family-centred care (FCC) was first described by Florence Blake (1954), who highlighted the crucial role of the family in children's care. Providing FCC means that clinicians incorporate into caregiving the knowledge that a family is a constant in children's lives and should therefore be involved in the caring process to promote the physical, emotional and psychological development of children and to achieve higher quality of care (Harrison, 2010). It is thus important to understand how families adapt when their children fall ill or become disabled (Harrison, 2010).

The core tenets of FCC include open communication and collaboration with clients and families in making decisions about care policies and services that will fit the family's needs and preferences in light of their cultural beliefs and

traditions. To support the family's strengths, both clients and families should be treated with dignity and respect, and each family's uniqueness should be understood (Arango, 2011). In the context of the child's disability, FCC strategies include understanding the needs of the whole family to encourage their potential and promote empowerment (Barbosa, Balieiro, & Pettengill, 2012). Knowledge about how life is experienced from an insider's perspective, i.e., from the perspective of children with SB and their family members, and how SB impacts their daily lives is a prerequisite for understanding what care and support is needed for these children and their families. Antle, Montgomery, and Stapleford (2009) emphasised the vital need for shared management of disability-related issues in children with SB in relation to client and family realities and to professional standards and goals.

1.4.3 SELF-PERCEIVED HEALTH VERSUS HEALTH-RELATED QUALITY OF LIFE

The World Health Organisation (WHO) introduced the basis for the definition of health in 1948 as a state of complete physical, mental and social well-being, not merely the absence of disease or infirmity (Karimi & Brazier, 2016). A few decades later, the concept of quality of life (QoL) emerged as an important outcome of health care (Elkinton, 1966). QoL is a broad and multidimensional construct covering all aspects of human life, including health, which makes it vague and difficult for use as a constant measure of health. Further, the lack of a unitary definition of QoL may lead to inconclusive findings (Bratt & Moons, 2015; Tennant, Pearce, Bythell, & Rankin, 2010).

Given that QoL is a subject of debate in the field of health and health care systems, a distinction has been made through the use of health-related quality of life (HRQoL) to improve patient health and well-being and to evaluate the effectiveness of health care (Lin, Lin, & Fan, 2013). In recent years, there has been growing interest in measuring patient reported outcomes (PRO), including HRQoL, that generally relies on the patient's subjective perception of health and functioning (Fayers & Machin, 2013). In WHO's International Classification of Functioning and Health (ICF) (World Health Organization, 2001), health and health-related domains are described as changes in body function and structure, i.e., what a person with a health condition can do in their usual environment (their level of performance). These domains are classified from the individual and societal perspectives by means of two lists: a list of body functions and structures, and a list of domains of activity and participation.

Currently, measures of HRQoL are usually more appropriately named measures of self-perceived health or self-reported health (Karimi & Brazier, 2016). In measuring self-perceived health in children, it is important to decide which approach to take, the broad multidimensional concept of health conditions and QoL, or the narrow definition of health-related functional dimensions, which include functional status and health perception (Wallander & Varni, 1998). This thesis used the latter approach to health and sought to measure physical, emotional, social and school functioning as designed in the 23-item Pediatric Quality of Life Inventory (PedsQL™ 4.0) Generic Core Scales (GCS) (Varni, Burwinkle, & Seid, 2005).

1.4.4 SENSE OF COHERENCE

The medical sociologist Aaron Antonovsky introduced salutogenic theory, which explores an individual's capacity to manage stress. Antonovsky (1979) tried to answer the (salutogenic) question: Why do some people, regardless of major stressful situations and severe hardships, stay healthy, while others do not? In response, he introduced the concept of sense of coherence (SOC) and claimed that life experiences shape one's SOC (Eriksson, Lindström, & Lilja, 2007). As the core of salutogenic theory, SOC may be viewed as part of resilience (Olsson, Hansson, Lundblad, & Cederblad, 2006). Antonovsky formulated a movement towards good health in terms of SOC, one which would enable people to manage tension and both identify and activate their social and psychological resources. The basis for the salutogenic perspective is an individual's childhood and social, historical and cultural context, in addition to genetic and environmental conditions. People with a low SOC have been found to experience more psychosocial stress compared to those with a higher SOC (Eriksson & Lindström, 2008).

Generalised resistance resources (GRR) is another important concept in salutogenic theory, one which provides individuals with sets of meaningful resources and coherent life; GRR is also about the power one has to fight against psychosocial stress (Antonovsky, 1979). Examples of GRR are cultural stability, coping strategies, engagement and social support. These resources are expected to develop a strong SOC, one which reflects the ability to cope with stressful situations. A person with a strong SOC has the ability to find appropriate solutions to specific stressors and can successfully resolve conflicts through adaptability.

Antonovsky claimed that the ways in which people view their lives have a positive influence on their health (Nilsson et al., 2010). Salutogenic theory gives a global orientation to view the world and the individual environment

with three components: (1) comprehensibility, which refers to the extent to which perceived stimuli are confronted as consistent, structured and clear; (2) manageability, or the extent to which one perceives that the resources at his or her disposal are adequate to meet life's demands; (3) meaningfulness, which refers to the extent to which one feels that life makes sense emotionally (Eriksson et al., 2007). The 13 items presented in the SOC-13 scale were built on the conceptualisation of these three components. Measuring SOC in adolescents with SB could reveal their ability to cope with their illness and disability in the Palestinian context.

1.4.5 STIGMATISATION

The concept of stigma, originally derived from the influential work of Erving Goffman, can be defined as 'an attribute that is deeply discrediting' (Goffman, 1963, p 3-4). This attribute can link a person to an undesirable stereotype and thereby cause disgrace: 'A stigmatized individual is the one who is disqualified from full social acceptance' (Goffman, 1963, p 3-4). Stigmatisation is often viewed as a social construct that identifies a group of people by attributing to them some distinguishing characteristics. It is also viewed as a specific case of prejudice (Jones, 1984). Stigma is composed of two fundamental elements: the recognition of a difference based on distinguishing characteristics, and the consequent devaluation of the stigmatised person (Major & O'brien, 2005).

According to Stuber, Meyer, and Link (2008) stigmatisation is a complex phenomenon that encompasses people's experiences, interactions between marginalised and non-marginalised groups, and broader structural and social phenomena, such as community practices, strategies, programmes, policies and power relations. In many cultures, people with apparent disabilities are often stigmatised when they do not meet normative expectations in social interaction. They are considered different and less valuable, and are labelled as such. At the same time, people with hidden disabilities might temporarily avoid stigmatisation (Kamenetsky, Dimakos, Aslemand, Saleh, & Ali-Mohammed, 2016). Given the negative traditional attitudes in Palestinian society towards disabilities and the potential blame placed on mothers, as described previously, stigmatisation should be considered a potential threat to the health and well-being of children with SB and their families.

2 RATIONALE

The literature provides knowledge about the burden experienced by children with SB and their family members that is relevant to the level of the impairment, the social context and the provision of health care services. A high quality of care is essential to reducing the impact of the disease on health and well-being. However, knowledge about how children with SB and their families manage their lives in countries plagued by political conflicts and poor socioeconomic conditions – like Palestine – is limited.

Similarly, studies on the lived experiences of children with SB and their mothers in Palestine are also limited. Furthermore, little is known about the self-perceived health and SOC in children with SB as compared with their healthy peers. Therefore, the reasons this research was conducted were to highlight the needs and challenges of children with SB and their mothers, to generate deeper understanding of living with SB and other disabilities in Palestine, and to augment nursing knowledge regarding this phenomenon in the broader context of long-term care.

Increased knowledge generated from this thesis may be beneficial in providing baseline data about children with SB and their families in Palestine. Further, the thesis may contribute to the development of health strategies that are compatible with the specific sociocultural milieu in Palestine.

3 AIM

The overall aim of this thesis was to describe the lived experiences of children with SB and their mothers in the specific social and cultural conditions of the West Bank, Palestine. Further, the thesis was intended to generate knowledge about the impact of SB on the self-perceived health and SOC of a group of children and adolescents with SB.

3.1 SPECIFIC AIMS

Study I: To illuminate mothers' lived experiences of having a child with SB in the West Bank, Palestine.

Study II: To illuminate the lived experiences of children with SB in the West Bank, Palestine.

Study III: (i) to measure self-perceived health in children with SB in the West Bank, Palestine, and compare them with a healthy reference group and (ii) to assess potential associations between self-perceived health and impairment levels in the SB group.

Study IV: (i) To assess self-perceived health and SOC in adolescents with SB in the West Bank in comparison to the healthy reference group, and (ii) to assess the potential associations between self-perceived health, SOC and impairment levels in the SB group.

4 PATIENTS AND METHODS

4.1 METHODOLOGICAL VIEWPOINT

This thesis employed a multi-method approach to understand the phenomena under study. A qualitative design (Study I and II) was chosen based on the assumption that the social context and sociocultural forces shape people's lived experiences, influencing health behaviour, perceptions and health outcomes.

The core methodological approach in the qualitative design (Study I and II) was the lifeworld perspective, which employs Husserl (1970) philosophy that all individuals live in a common world and share experiences. An orientation towards the complex world and lived experiences of children with SB and their mothers was the central concern in these studies. Understanding was gained through interpretation of the subjective experiences of children with SB and their mothers living in the West Bank, Palestine.

Narrative interviews were conducted in Study I and II to collect data from the mothers and their children, i.e., to elicit views, thoughts, perceptions and feelings about their lived experiences. Narrative interviews are considered in-depth, unstructured tools, with specific features that arise from the life stories of both the participants and the cross-examined situational context. The narrative is a traditional way of communicating meaningful content, from which experiences can be transmitted. The main goal of narrative interviews is to encourage and stimulate the participants to tell the researcher something about important events in their lives and about the social context (Muylaert, Sarubbi Jr, Gallo, Neto, & Reis, 2014). This requires the researcher's skills in guiding the interview and in formulating the interview questions. An important collaborative feature appears within this method, since the story emerges from the interaction, exchange and dialogue between the researcher and the participants. However, the influence of the researcher in narrative interviews should be minimal, based on the idea of reconstructing social events from the point of view of the participants.

Narrative interviews assumes that the perspective of the participants is best revealed when they use spontaneous language. This facilitates understanding of people's subjectivities in the context of the events that they bring to mind and that they convey in the inter-subjective context of the interview, using their own expressions (Hollway & Jefferson, 2008). Narratives are not open to evidence and are considered to be representations and interpretations of the

world as lived by the participants and, therefore, cannot be judged as true or false (Muylaert et al., 2014).

The phenomenological-hermeneutical method was chosen in Study I and II. This method accommodates the philosophical assumption of qualitative studies and follows the scientific process of understanding and interpreting the data. Epistemologically, phenomenological approaches are based on a paradigm of personal knowledge and subjectivity and emphasise the importance of understanding subjective experiences. According to Lindseth and Norberg (2004), to understand human consciousness and actions, phenomenology must be supplemented by hermeneutics. The phenomenological-hermeneutical method is an attempt to disclose the world as experienced by the subject through their lifeworld stories and to generate interpretations of a phenomenon through the use of the hermeneutic cycle in the analysis. Thus, interpretation of a text is necessary to understand its meaning and reveal the phenomenon in question (Lindseth & Norberg, 2004).

In the quantitative studies (Study III and IV), self-reported data were gathered from children and adolescents with standardised health measures – PedsQL™ 4.0 and SOC-13 – to understand and complement how they experience their life situations and perceive their health and functioning. For the younger children, self-perceived health was measured with the PedsQL™ 4.0 GCS inventory in Study III; whereas in Study IV, self-perceived health and SOC among adolescents were assessed with the PedsQL™ 4.0 GCS inventory and the SOC-13 scale.

4.2 RESEARCH DESIGN

To capture, explain and understand the phenomenon of living with SB in Palestine, a multi-method approach was used that incorporated both qualitative and quantitative data. The four studies were descriptive and included the qualitative phenomenological-hermeneutical approach and a quantitative cross-sectional design (see Table 1).

Table 1. Overview of the research design for all studies in the thesis

Study	Design	Data collection	Participants	Data analysis
I	Qualitative lifeworld perspective	Narrative interviews	20 mothers of children with SB Aged 29–62 years	Phenomenological -hermeneutical method
II	Qualitative lifeworld perspective	Narrative interviews	10 children with SB Aged 7–18 years	Phenomenological -hermeneutical method
III	Cross-sectional	Questionnaire PedsQL™ 4.0	50 children with SB 150 healthy children Aged 8–12 years	Descriptive and comparative statistics
IV	Cross-sectional	Questionnaire PedsQL™ 4.0 SOC-13	50 adolescents with SB 150 healthy adolescents Aged 13–18 years	Descriptive and comparative statistics

SB = Spina Bifida. PedsQL = Pediatric quality of life inventory. SOC = Sense of coherence.

4.3 SETTINGS AND PARTICIPANTS

4.3.1 STUDY I AND II

The participants were recruited purposefully from the three largest rehabilitation centres in the north, middle and south of the West Bank, Palestine, and were contacted by the nurses working in these centres. In total, 20 mothers of children with SB were selected in Study I, while 10 children with SB (six males, four females) were selected in Study II. The inclusion criterion for mothers was that they had a child with SB. The strategy was to include mothers of different ages and places of residence, and also to cover a wide range of SB forms and severities in their children. The inclusion criteria for children in Study II were that they were aged 7–18 years and had good communication abilities that makes it possible for them to contribute to the data. Children were selected purposefully with variations in gender and places of residence (different districts in the West Bank) as well as a wide range of SB forms and consequences. Exclusion criteria were a documented history of learning disabilities or limited cognitive function. None of the initially contacted mothers and children declined to participate in the studies.

4.3.2 STUDY III AND IV

The samples in Study III and IV were convenient and involved 200 participants (50 in the SB group and 150 in the reference group), respectively. The participants in Study III were aged 8–12 years, while those in Study IV were aged 13–18 years. All children and adolescents with SB were selected from the three largest rehabilitation centres in the West Bank, Palestine. The number of selected children and adolescents in Study III and IV were, respectively, 23/26 from the south (Hebron district), 9/11 from the middle (Ramallah district) and 18/13 from the north (Nablus district) of the West Bank. The inclusion criteria for children and adolescents in the SB group were a confirmed diagnosis of SB and age within the previously identified groups, while the exclusion criteria were other medical disorders or cognitive deficits. Nurses working in the rehabilitation centres identified eligible children and adolescents with SB and contacted their parents to ask for their participation. The reference group of healthy children and adolescents were recruited from different primary and high schools located in different districts in the West Bank. The number of selected children in Study III was 65 from Hebron, 45 from Ramallah and 40 from Nablus; similar numbers of adolescents were selected for Study IV. The heads of the schools contacted the children and adolescents as well as their parents in Study III and IV. In Study III, nurses identified 54 children as eligible, yet four children declined to participate because they were sick on the dates on which the study was conducted. In Study IV, 55 adolescents with SB were identified, two of whom declined to participate without giving a reason, while three were withdrawn from the study due to hospital admission for shunt repair at the time of their scheduled visit to the centres. Thus, 50 participants ultimately took part in Study IV.

Table 2. Demographic characteristics of the participants in the four studies

Study		Study I	Study II	Study III		Study IV	
Participants		Mothers of children with SB	Children with SB	Children with SB	Reference group	Adolescents with SB	Reference group
Number		20	10	50	150	50	150
Age group		27-65	7-18	8-12	8-12	13-18	13-18
Age (mean)		41.8	11.1	10	11	14.6	14.5
Gender of children	Male (%)	13 (65)	6 (60)	27 (54)	76 (51)	23 (46)	72 (48)
	Female (%)	7 (35)	4 (40)	23 (46)	74 (49)	27 (54)	78 (52)

Place of residency	Urban (%)	5 (25)	3 (30)	8 (16)	108 (72)	16 (32)	63 (42)
	Rural (%)	7 (35)	4 (40)	27 (54)	25 (17)	22 (44)	40 (27)
	Camp (%)	8 (4)	3 (30)	15 (30)	17 (11)	12 (24)	47 (31)
Child disability	Paralyzed (%)	11 (55)	6 (60)	24 (48)		21 (42)	
	Can walk (%)	9 (45)	4 (40)	26 (52)		29 (58)	
	Hydrocephaly (%)	9 (45)	8 (80)	26 (52)		22 (44)	
	No hydrocephaly (%)	11 (55)	2 (20)	24 (48)		28 (56)	
	Bladder dysfunction (%)		10 (100)	50 (100)		50 (100)	
	Bowel dysfunction (%)		10 (100)	50 (100)		50 (100)	

4.4 DATA COLLECTION

4.4.1 STUDY I AND II

Data were collected through narrative interviews with participants in their homes. The interviews were conducted in private, and the participants were encouraged to speak freely to promote narration. All interviews were audio-recorded and lasted for 45 to 80 minutes (mean = 60 min.) in Study I, and 30 to 40 minutes (mean = 35 min.) in Study II. The interviews started by asking one opening question about the participants' daily life experiences with SB. Follow-up questions were asked when needed. Data were collected in Arabic, transcribed verbatim, and then translated into English.

The participants felt comfortable in their home environments, and privacy was maintained as much as possible, with distractions, such as interruptions, noise or other visible activity, kept to a minimum. Mothers were encouraged to speak freely about their experiences of having a child with SB and were asked broad, open-ended questions. The opening question was, "Tell me about your experience of having a child with spina bifida." Additional follow-up questions, such as "Can you tell me more about . . ." and "How did you feel when . . .," were posed to clarify statements and to confirm the interviewer's understanding. The children were asked one broad question: "Could you tell me about your experience with spina bifida?" Follow-up questions were asked that were open-ended, specific and clear. Throughout the study process, the guidelines for communicating with children (Hockenberry & Wilson, 2003) were followed to protect the participants, the researcher and the data. These included giving the participants the opportunity to talk without threat, listening to them, avoiding sudden gestures and asking clear and specific questions to allow them to express their concerns and fears. After the interview, the interviewer made sure that the participants were not distressed and assessed whether emotional support was needed; the researcher also asked the participants about how they experienced the interview situation and about their thoughts and feelings – most of the interviewed mothers and children said that they found it positive to share their experiences and to have someone listen to their thoughts. They also felt that they had gained new insights into their own situation. Appendix I includes the interview guides for Studies I and II.

4.4.2 STUDY III AND IV

The PedsQL™ 4.0 GCS, Arabic version, was used to collect the data in Study III and IV, while the SOC-13, Arabic version, was used in Study IV only. The PedsQL™ 4.0 GCS consists of 23 items on a 5-point Likert scale. It measures two dimensions of health: a physical health summary score (Physical

Functioning Subscale, eight items) and a psychosocial health summary score (Emotional, Social and School Functioning Subscales, including five items for each subscale). The instrument is applicable to children and adolescents aged 5–18 years in healthy school and community populations as well as in paediatric populations with acute and chronic health conditions (Varni et al., 2005). The Arabic version of the PedsQL™ 4.0 GCS is reliable and valid for investigating children's self-perceived health in Arab countries. It possesses psychometric properties within the range considered acceptable in other cross-cultural research studies (Abdul-Rasoul, AlOtaibi, AlMahdi, & AlKandari, 2012). The SOC-13, Arabic version, is applicable to healthy school and community populations as well as to paediatric populations with acute and chronic health conditions (Eriksson & Lindström, 2005). It consists of 13 items reflecting three dimensions: meaningfulness (four items), comprehensibility (five items) and manageability (four items). Appendix III includes the PedsQL™ 4.0 GCS and SOC-13. The SOC cut-off points in the present study were arbitrarily divided on the basis of the previous studies (Mendel, Bergenius, & Langius, 2001): scores of 35–60 were considered low; scores of 61–75 were considered moderate; and scores ≥ 76 were considered strong.

For Studies III and IV, the participants from the two groups and their parents were first informed about the study; prior to completing the forms, the procedure, confidentiality and the optional nature of participation were made known to the participants.

The children and adolescents (Studies III and IV) completed the questionnaire responses on their own with no assistance, and there were no cases in which a parent proxy was used in place of an actual subject response. Qualified and trained nurses working at the rehabilitation centres were present with the children with SB during the course of completing the forms, which took approximately 10–15 minutes, while I, or a research colleague, accompanied the reference groups in schools. For children who demonstrated or indicated that they had difficulty understanding the questions, the items were read aloud and explained, and time was provided for the child to record his or her answers.

4.5 DATA ANALYSIS

4.5.1 STUDY I AND II

A phenomenological hermeneutical approach, inspired by Ricoeur's theory of interpretation (Paul Ricoeur, 1976) and described by Lindseth and Norberg (2004) was used in Studies I and II. According to (Paul Ricoeur, 1981), phenomenological and hermeneutical approaches are mutually interrelated

such that phenomenology has an intermediate nature that hermeneutics discloses, and hermeneutics itself cannot be constituted without the phenomenological condition. This means that the process of interpretation requires both understanding and explanation in order to construct knowledge that is valid and can be incorporated to the on going discourse.

This qualitative approach was chosen to explore the phenomenon of living with SB and being a mother of a child with SB, as described by the mothers and children. To gain in-depth knowledge of this phenomenon, lived experiences should be both understood and explained in a scientific way. Accordingly, the theory of interpretation shifts between two different approaches to the text – subjective, in order to understand the text, and objective, in order to explain it by employing various structural analyses (Paul Ricoeur, 1981).

According to Lindseth and Norberg (2004) the phenomenological-hermeneutical approach involves understanding and interpreting the meaning of a text through naïve reading, structural analysis and comprehensive understanding. Making the meaning visible requires interaction between the researcher and the text in a circular, stepwise process, shifting back and forth between understanding and interpreting to reach a comprehensive understanding of the phenomenon under study. Naïve understanding begins by reading the entire text several times to obtain an immediate sense and broad picture of its content. In the structural analysis, each section is read carefully. Then, meaning units relevant to the aim are identified, abstracted and critically reviewed in relation to the naïve understanding. Meanwhile, categories, patterns and relations between meaning units were explored to form subthemes and themes and to formulate the main themes. In this study, preliminary understandings were discussed before the analysis and were constantly reconsidered and reflected upon during the entire data analysis and interpretation process.

Table 3. Example of the structural analysis in study I

Meaning Unit	Subtheme	Theme
It is a misery, I am always nervous, I can't tolerate myself, I can't walk as other children	Resentment against disability	Experiencing negative self-concept
I'm totally dependent, I can't manage my incontinence, my mother help me to deal with this, even at school, but when I'm older who will help me?	Powerlessness and dependency	Experiencing negative self-concept
I do not use a wheel chair, but I'm paralyzed, I cannot run like other children. I get angry when they laugh at me or make fun of me. Sometimes I cry, and sometimes I want to hit them with my crutches	Struggle with being different	Experiencing negative self-concept

4.5.2 STUDY III AND IV

The data were analysed using SPSS statistics software (IBM SPSS Statistics 24, 2016). Descriptive statistics (frequencies, percentages, means and standard deviations) were used in Study III and IV to summarise the demographic characteristics of the study population in both groups. In Study III, the Mann-Whitney non-parametric U test was used to compare the total PedsQL™ 4.0 scores between the group of children with SB and the reference group. The same test was also used in Study IV to compare the total scores of PedsQL 4.0 and SOC between the adolescent group with SB and the reference group. The associations between level of impairment and self-perceived health in children and adolescents with SB were determined through the use of a Spearman's rank association (r_s) test. The same association test was used to investigate the association between level of impairment and SOC in adolescents in study IV.

4.6 ETHICAL CONSIDERATIONS

The four studies in this thesis were planned in accordance with the parameters of the Declaration of Helsinki (World Medical Association 2013). The Universal Human Rights Declaration (Assembly, 1948), also guided the studies, as well as the UN Convention on the Rights of the Children (Unicef, 1989). Local permissions to conduct the studies were obtained from the Palestinian Health Authority, and approval for conducting the research was obtained from the Research Ethical Review Committee at Al-Quds University, Palestine.

The scope and participants for this thesis necessarily expose vulnerabilities, as the population investigated in Studies II, III and IV were children and adolescents. Ethical considerations required for studying human beings were therefore carefully observed. Care was exercised in ensuring that the four commonly accepted principles of health care ethics derived from Beauchamp and Childress (2008) – respect for autonomy, nonmaleficence, beneficence and justice – guided the data collection and presentation of the thesis results.

The participants were informed that participation was voluntary and that all information would be treated confidentially. Their rights to withdraw from the study at any time were emphasised, and they all signed consent forms and agreed to audio recording of the interviews. In Study I, the mothers signed the consent, while parents of the children in Studies II, III and IV were asked to sign the consent after being provided with written information about the study. The consent of the children and adolescents who participated was received before asking for written parental consent.

Children and adolescents are vulnerable groups and demand special precautions to avoid violating their rights and integrity. Assuring confidentiality, respect, protection and empowerment of the children is required throughout the research process. Further, ethical considerations concerning risks of emotional harm in raising issues related to sensitive matters were considered. As an interview may evoke painful memories and emotions, assessment of the participants' needs for emotional support was undertaken. Emotional support was given to children once needed, and a psychotherapist at the rehabilitation centre was available if needed. Children were reminded of their rights to stop or withdraw from the study at any time. However, none of the participants required professional help, and I found that my experience as a paediatric nurse was sufficient to provide support during the emergence of memories and emotions. During emotional events, I stopped the interviews and gave the participants time to cry or otherwise express significant emotions.

5 RESULTS

The main findings from the four studies are summarised and presented, study by study, in the following sections.

5.1 STUDY I

In this study, the mothers' stories reflected how they faced challenges in their life situations and how they were struggling to promote the health and well-being of their children with SB and other family members. The structural analysis and interpretation were dominated by the following main theme: 'From feeling broken to looking beyond broken'. Four interwoven themes were formulated in condensed descriptions to assemble the subthemes and convey the meaning of the mothers' experiences: living with constant anxiety, living with uncertainty, living with a burden, and living with a difficult life situation (Table 4).

The mothers in this study felt stigmatised for giving birth to a child with SB, which affected their emotional status and led to a state of constant anxiety. The most prominent hardship situations were experienced at the time of diagnosis and just after the child's birth, as the mothers lived with fears, worries, blame and guilt. The mothers also lived with uncertainty about their child's health and future life. Their limited knowledge about SB increased their sense of burden and uncertainty. They were overwhelmed with the critical health conditions and caring responsibilities concerning their child. Mothers often experienced social isolation and the need for emotional support. They found this support within their families, while consolation was found in spiritual and religious beliefs.

5.2 STUDY II

The structural analysis of the children's narratives revealed their lived experiences, as illustrated in the main theme: 'vulnerability and suffering due to social exclusion and stigma'. Three themes were formulated in condensed descriptions to assemble the subthemes and convey the meaning of the children's experiences: experiencing negative self-concept, experiencing vulnerability, and obtaining a sense of security (Table 5). Children shared their reflections on their self-concept, which were primarily negative. They described their physical appearance, personal attributes and sense of self through a series of painful emotions. Feeling dependent was one of the main difficulties reported by the adolescents, but it was not considered a problem for

the young children. Some adolescents reported facing difficulties acquiring independence by managing the clean intermittent catheterization (CIC) and other health routines by themselves. These threats to self-management and independence intensified their emotional suffering and negatively affected their self-concept. They were more occupied than the younger children with thoughts about the future, and they reported concerns about their education, future careers, work and marriage.

The children's experiences of vulnerability were related to negative social attitudes and environmental barriers, especially at school, which influenced their feelings about living with stigmatisation. Living with limitations was illustrated by the children's problems with accessing buildings and streets, which were not disability-friendly. The children found it difficult to reach school or places of recreation appropriate for their age on their own. However, family and religious beliefs were described as sources of support that promoted the children's feelings of security and belonging.

Table 4. Overview of the main theme, themes and sub-themes in the study of Palestinian mothers' lived experience of having a child with SB (Study I).

Main theme <i>From feeling broken to looking beyond broken</i>				
Themes	Living with constant anxiety	Living with uncertainty	Living with a burden	Living with a difficult situation
Subthemes	Feeling fears and worries. Feeling blamed and guilty	Uncertainty about the child's health. Uncertainty about the child's future	Constantly meeting challenges. Going it alone with the caregiver burden	Experiencing social isolation. Being in need of emotional support Finding spiritual consolation

Table 5. Overview of the main theme, themes and subthemes in the study, children's descriptions of their experience of living with SB (Study II).

Main Theme <i>Vulnerability and suffering due to social exclusion and stigma</i>			
Themes	Experiencing negative self-concept	Experiencing vulnerability	Obtaining a sense of security
Subthemes	Resentment against disability Powerlessness and dependency Struggle with being different	Living with stigmatization Living with limitations Risk to body and self	Belonging within the family Belonging with peers

5.3 STUDY III

This quantitative study assessed self-perceived health in children with SB in comparison with a group of healthy children. Demographic data were collected from both groups, as were their answers to the 23 items on the PedsQL™ 4.0. The mean age for the SB group was 10.0 (SD \pm 1) years; for the reference group, it was 11.0 (SD \pm 1) years. The total mean of the PedsQL™ 4.0 score scale was 45.5 (\pm 14.5) in the SB group and 80.8 (\pm 12.8) in the reference group. Self-perceived health was found to be significantly lower for the 50 children in the SB group than it was for the 150 children in the reference group ($p < 0.001$) with regard to each dimension: physical health summary score ($p < 0.001$) and psychosocial health summary score ($p < 0.001$). For children with SB, scores varied among the PedsQL scales. Social functioning had the lowest mean score (30.5), followed by physical functioning (37.5), emotional functioning (56.0) and school functioning (62.6), while the range for all the PedsQL scale scores in the reference group was between 71.7 and 84.5 (Table 6).

No associations were found between demographic characteristics of age, gender or residency category and self-perceived health in either group. Higher levels of impairment in mobility and hydrocephaly with shunt were negatively associated with self-perceived health with respect to total PedsQL score: r_s -0.65 ($p < 0.001$), r_s -0.53 ($p < 0.001$), respectively. Unexpectedly, no correlations were seen between the levels of bladder or bowel dysfunction and self-perceived physical or psychosocial health, despite the generally low reported scores for the dysfunction.

Table 6. Comparison of Self perceived health between children with SB and healthy children (Study III).

	SB Group (n=50)	Reference group (n=150)	
PedsQL 4.0	Mean (\pm SD)	Mean (\pm SD)	p-value**
Total scale score	45.5 (\pm 14.5)	80.8 (\pm 12.8)	< 0.001
Physical health summary score	37.5 (\pm 16.6)	84.0 (\pm 13.3)	< 0.001
Psychosocial health summary score*	49.7 (\pm 15.5)	79.0 (\pm 14.3)	< 0.001
Emotional Functioning scale	56.0 (\pm 17.2)	71.7 (\pm 19.1)	< 0.001
Social Functioning scale	30.5 (\pm 20.8)	84.5 (\pm 18.4)	< 0.001
School Functioning scale	62.6 (\pm 17.8)	80.9 (\pm 18)	< 0.001

SD = standard deviation. PedsQL= Pediatric quality of life inventory

*The psychosocial health summary score is calculated as the sum of the items divided by the number of items answered on the emotional functioning, social functioning and school functioning subscales.

**Statistical comparisons were conducted with the Mann-Whitney U test

5.4 STUDY IV

This quantitative study assessed self-perceived health and SOC in adolescents with SB in comparison with a group of healthy adolescents. Demographic data were collected from both groups, as were their answers to the 23 items in the PedsQL™ 4.0 and the 13 items in the SOC. The mean age for the SB study group was 14.6 years ($SD \pm 1.6$); for the reference group it was 14.5 years ($SD \pm 1.7$). The 50 adolescents with SB perceived their health to be lower than that of the 150 adolescents in the reference group in terms of total PedsQL score ($p < 0.001$) and with regard to each dimension: physical health summary score ($p < 0.001$) and psychosocial health summary score ($p < 0.001$).

The total PedsQL™ 4.0 score scale was 54 ($SD \pm 18$) in the SB group and 82 ($SD \pm 11$) in the reference group. In the SB group, physical functioning had the lowest mean score (42), followed by the social mean score (45), emotional functioning (62) and school functioning (71), while the range for all the

PedsQL scale scores in the reference group was between 70 and 91; the lowest score was for emotional functioning, while the highest was for social functioning (Table 7)

The total SOC score was 47 ($SD \pm 5$) in the SB group and 55 ($SD \pm 14$) in the reference group. Differences between the two groups were also found regarding two aspects of SOC: meaningfulness ($p < 0.001$) and comprehensibility ($p < 0.02$). No differences were seen with regard to manageability, which had a p -value of 0.36 (Table 7). The SOC score was positively associated with increased scores for self-perceived health in both groups.

Self-perceived health in the SB group was negatively associated with the level of impairment in the total score scale. Impairment in mobility was associated with a value of r_s -61 ($p < 0.01$); hydrocephaly was associated with a value of r_s -60 ($p < 0.01$); bowel function was associated with a value of r_s -38 ($p < 0.01$); bladder function was associated with a value of r_s -17 ($p < 0.05$). A negative association was also found between SOC in adolescents with SB and their impairment level. Bladder incontinence was associated with a value of r_s -42 ($p < 0.01$), hydrocephaly r_s -32 ($p < 0.01$), impairment in mobility r_s -28 ($p < 0.05$), and impairment in bowel function r_s -27 ($p < 0.05$).

Table 7. Comparison of self-perceived health and SOC between adolescents with SB and healthy adolescents (Study IV)

	Spina bifida group (n=50)	Reference group (n=150)	
PedsQL 4.0	Mean (± SD)	Mean (± SD)	p-value***
Total scale score	54 (±18)	82 (± 11)	<0.001
Physical health summary score	42 (±19)	86 (±13)	<0.001
Psychosocial health summary score*	58 (± 19)	81(±12)	0.01
Emotional Functioning scale	60 (± 25)	70 (± 19)	<0.001
Social Functioning scale	45 (±27)	91(± 13)	0.003
School Functioning scale	71 (± 23)	81 (± 18)	<0.001
Sense of Coherence			
Total scale score**	47 (±5)	55 (±14)	<0.001
Meaning fullness, 4 items	14 (±3)	19 (±6)	<0.001
Manageability, 4 items	15 (±2)	16 (±5)	0.36
Comprehensibility, 5 items	18 (±2)	20 (±8)	0.022

SD = Standard deviation. PedsQL = Pediatric quality of life inventory. SOC = Sense of coherence.

* The psychosocial health summary score is calculated as the sum of the items divided by the number of items answered on the emotional functioning, social functioning and school functioning scales.

** SOC total scale score is calculated as the sum of the items divided by the number of items answered on the meaningfulness 4 items, manageability 4 items and comprehensibility 5 items scales.

*** Statistical comparisons were conducted using the Mann–Whitney U test.

6 DISCUSSION

In this thesis, a qualitative method was used to illuminate the phenomenon of the lived experiences of children with SB and their mothers in the West Bank, Palestine. Further, a quantitative study was conducted to assess self-perceived health and SOC among children and adolescents with SB in comparison with healthy peers.

The interview data in the qualitative studies (Study I and II) provided a range of shared insights as experienced by the participants from the two studies. Children with SB and their mothers explored SB-related psychological distress, challenges in social life and the essential need for care and support. The feelings, behaviours, social interactions and health conditions of the mothers and children were illuminated through the interpretation of their narratives.

The results in Study I were summarised into one main theme, ‘from feeling broken to looking beyond broken’. This means that a sense of broken feelings and emotional suffering influenced the mothers for a long time before they were able to tap into their intrinsic resilient capabilities and decide to look beyond broken. This result is consistent with a study by Monsen (1999) in which the lived experiences of mothers when parenting a child with SB were described as ‘living worried: staying in the struggle’ Monsen (1999, p. 161) while resilience was seldom reported.

In Study I, the mothers experienced internal stress at the time of the child’s birth, related to the negative social attitudes and stigma of giving birth to a child with abnormalities. They lived with constant anxiety and uncertainty for long periods before they gained the ability to cope with the child’s conditions and to focus on the child’s health and the future life of the family. The profound impact on mothers’ psychosocial health and QoL of caring for a child with SB was described in the longitudinal study of Brekke, Früh, Kvarme, and Holmstrøm (2017) which was conducted in Norway and which indicated that the burden of care falls particularly on the mother.

In Palestine, it is well known that women are the caretakers of family members – the sick, older people and people with disabilities. This predefined care-taking role is sex-linked and is dictated by a patriarchal society (Giacaman, 2001). However, mothers of children with SB often feel restricted by their parenting role, less competent as a parent and socially isolated. They also

experience higher stress levels than fathers do (Vermaes, Janssens, Mullaart, Vinck, & Gerris, 2008).

The mothers' narratives (Study I) revealed the burden of managing their children's care. The mothers felt lonely with the caregiving burden. They faced difficulties in meeting the emerging needs and challenges of a sick child, the demands of other family members, the economic burden and the deficient health and social resources in Palestine. This burden was also associated with the consequences of SB and the lack of knowledge about SB care. Further, political factors added to the children's and their families' burdens, by restricting their ability to reach hospitals that provide advanced surgeries and devices to maintain the child's health. Furthermore, the CBR models of care call for increasing the mothers' burdens (Giacaman et al., 2011), because the responsibility of caring for the child, although it is family-based, in reality translates primarily into a responsibility of the mother. Rehabilitation in the home means that the mother needs to learn yet more skills, to work more with the child at home and to deal with the child's daily health and problems. The mothers and families found that integrating the child into schools and societies, in terms of time and energy, let alone the financial aspects, is very difficult to achieve.

The findings in Study I and II showed how stigma and discrimination against disabled people represent serious psychosocial stressors for both mothers and children. The negative sociocultural attitudes prevalent in Palestinian society place blame on the mothers, viewing them as being responsible for their child's abnormality. Consequently, the mothers lived with constant anxiety and worried about social acceptance and loss of their reputation by giving birth to a child with SB. The mothers' worries in relation to family acceptance and social exclusion are common in other Middle Eastern countries that perceive disability as a stigma (Hasnain et al., 2008). The impact of stigma on the mothers was similar to that experienced by mothers in the United Arab Emirates, according to Crabtree (2007), who explored its potential threat to marital stability and social standing, which in turn impacts the social inclusion of disabled children.

The children with SB (Study II) were also struggling with stigma, particularly social prejudice and strained social interactions with peers. The children expressed feelings of embarrassment, secrecy and shame. The findings in Study II were summarised in the main theme, 'vulnerability and suffering due to social exclusion and stigma'. This means that the children were burdened by the complexity of SB and its consequences, as well as by the associated discrimination and social exclusion, which affected their physical and

psychosocial health and well-being. These findings are in line with those produced by previous studies that highlighted the impact of stigma on children with disabilities in different contexts (Cocks, 2008; Connors & Stalker, 2007; Jahoda, Wilson, Stalker, & Cairney, 2010). Although children with disabilities in other Arab Muslim countries, as well as in developing and low-income countries, suffer a similar impact linked to stigma and marginalisation (Hasnain et al., 2008), the adversity faced by our participants was more complicated. Children with birth defects in developing countries, like Palestine, encounter unique illness- and disability-related stressors (WHO, 2011). They might not have the same opportunities that children have in other countries for habilitation, training, education and counselling, since Palestinian children are burdened by a protracted political conflict that has overwhelmed the country's infrastructure and strained its economy, health care system, rehabilitation services and service providers (Giacaman et al., 2009).

An important finding in this thesis concerns children's fears of the stigma linked to incontinence and the use of CIC in school (Study II). Their fears of prejudice, rejection and exclusion have led them to hide their incontinence, to pretend to use bathrooms as their healthy peers do, and to avoid mixing with others at school. However, the participants' attempts to hide and/or postpone the use of CIC in school may be related to their lack of knowledge about CIC and its importance in preserving renal function. It is therefore vital to discuss these issues with the children and their families at a young age and to train them in how to manage CIC early on. Doing so would benefit the children's health and reduce renal complications (Fischer, Church, Lyons, & McPherson, 2015). A study by Fischer et al. (2015) found that children who can control their bladders with only minimal accidents have greater independence and more opportunities for social participation than children who cannot. Further, such control can enable them to feel more comfortable, less stigmatised and more dignified (Fischer et al., 2015; Sawin, Bellin, Roux, Buran, & Brei, 2009).

The children's negative self-concept, as described in Study II, is an important and common issue among children with SB. A meta-analysis by Shields, Taylor, and Dodd (2008) related poor self-concepts among children with SB to symptoms characteristic of defects, such as incontinence, paralysis and deformities, which can impact their functioning and interactions with others. Shields et al. (2008) also reported that children with SB scored significantly lower than their peers in the domains of global self-worth, physical appearance and social acceptance.

The feelings of dependency expressed by the children in Study II are congruent with findings by Holmbeck et al. (2003) which revealed that children with SB are more dependent on adults and are less likely to make independent decisions than healthy children. Interestingly, independence in Study II was perceived differently between the children and the adolescents. The adolescents were struggling to gain autonomy and were more occupied with their thoughts about the future than the younger children, who showed satisfactory feelings about being dependent. These differences can be linked to their stage of development. Achieving independence is the most important aspect of the adolescent period and is accompanied by the emergence of other age-related milestones, including self-awareness, the identification of various needs and the acquisition of abstract thinking (Helseth & Misvær, 2010).

In this thesis, both mothers and children (Study I and II) described the family relations, religious beliefs and spirituality as the most important sources of support; these sources helped them regain some of their psychosocial well-being and strength. Children felt attached, safe and secure in their home environment. Family assistance and support mitigated the mothers' feelings of grief, enhanced their psychosocial well-being, fostered their new caring role and allowed them to face negative social attitudes and cope with their child's difficult condition. The previously mentioned study from the Middle East emphasised that mothers' and children's ability to cope with disability-related conditions is connected to the supportive role provided by the family and to the influence of religious beliefs (Crabtree, 2007).

The systematic literature review conducted by Bakaniene et al. (2016) suggested that family functioning, social support and resilience are significant factors that can ameliorate the impact of SB on children's health. These results agree with those generated by Antle et al. (2009); in their study, the importance of the FCC model of practice in paediatric rehabilitation for strengthening and empowering children with SB and their families was emphasised. FCC is usually offered by a multidisciplinary health care team that works to promote physical, emotional and psychosocial development in families. It is essential that this approach ensures that the services provided are appropriate for the family's culture and traditions, and that support is provided to sustain the resources within the family unit.

Both the mothers and the children in Studies I and II showed a sense of spirituality that seemed to help them to improve their abilities to deal with the complex conditions of SB. It is apparent that patients suffering from chronic illness and disability, and facing uncertainty as to the course of their lives, could potentially benefit from spiritual reflection (Naghi, Philip, Phan,

Cleenewerck, & Schwarz, 2012). Indeed, spiritual exploration and practices in daily life are often connected with religion, which provides a structured environment for spirituality. Religions are based on the existence of a higher power and the healing properties of spiritual experiences (David, 2008). The mothers and children in Studies I and II possessed awareness and appreciation of something beyond human faith – belief in God or a higher power that is in control. This discovery of a meaning in life through spirituality afforded the mothers and children a source of support.

This thesis highlights the multitude of issues in the Palestinian context that impact the health and psychosocial conditions of individuals and that burden children with SB, their mothers, and their family members. That said, according to Study I and II, children with SB and their mothers benefit from family support and religious beliefs, and yet there are other contextual issues in Palestine that complicate their experiences. These issues include economic hardship, lack of social resources and rehabilitation services, environmental barriers, unattainable inclusive education, restricted movement and the consequences of ongoing political instability in Palestine. Many of these factors were also highlighted in a study by Marie, Hannigan, and Jones (2016), who emphasised their impact on health and service needs in Palestine.

Although the PA has been committed to promoting the social rights of children with disabilities, as outlined by the UN convention and the Millennium Development Goals for persons with disabilities (UNICEF, 2016), the transition to a rights-based approach is very slow. Normal access to health services, social support, education and work are often unattainable (Burton et al., 2013). Despite the establishment of a comprehensive body of legislation, policies and CBR programmes that are consistent with social justice, there continues to be a gap between laws, policies and practice. The implementation gap might be related to stigma and the negative cultural attitudes towards disability, the political consequences and the above-mentioned multitude of issues in Palestine.

When assessing self-perceived health among children and adolescents in Studies III and IV, a significant difference was found between the study groups and the reference groups in all dimensions of the PedsQL. These results are consistent with previous studies (Johansen, Dammann, Andresen, & Fagerland, 2013; Müller Godeffroy et al., 2008), as children with SB in these studies perceived their HRQoL as lower than did their healthy peers.

Data from a previous literature review (Bakaniene et al., 2016) showed consensus in the significant impact of physical factors on the children's health

and functioning, while the reported findings about the psychosocial factors have been inconsistent. Although Copp et al. (2015) considered physical, neurocognitive and social functioning to be important factors in lower HRQoL among children with SB, Johnson, Nilsson, and Adolfsson (2015) related the greatest reduction to physical functioning and the smallest reduction to emotional functioning, while Jenkinson et al. (2011) suggested other predictors of HRQoL, including social class, levels of pain, parenting stress and family functioning.

However, children in this thesis (Studies III and IV) had low scores on the social functioning subscale; although the adolescents' (Study IV) lowest scores were not for social functioning, unlike the children (Study III), they still reported social scores that were very low. The lower scores for social functioning in Studies III and IV might be partly related to the negative social attitudes and environmental obstacles in Palestine that were explored by the children in study II. Kaur et al. (2016) also claimed that social and environmental aspects in Palestine could exacerbate the social exclusion of children with disabilities.

Moreover, the reported scores on the total PedsQL scale in Studies III and IV, among children with SB, were lower than the findings from self-report studies conducted in other contexts (Abresch, McDonald, Widman, McGinnis, & Hickey, 2007; Ramachandra, Palazzi, Skalsky, Marietti, & Chiang, 2013). Although SB can profoundly impact the physical and psychosocial functioning of affected children, the contextual factors can also have an important impact (Bannink et al., 2016). These results might be partly related to the multitude of factors in the Palestinian context that influence individuals' lives in general, including socioeconomic status, political issues, the provision of care and the lack of resources. Studies have emphasised the impact of these factors in the deterioration of the psychosocial health and social adjustment of children with SB (Bannink et al., 2016).

In Studies III and IV, impairment in mobility and shunted hydrocephalus were both inversely associated with self-perceived health, which is in line with a previous study by Ramachandra et al. (2013). The positive impact of mobility independence on self-perceived health and QoL has also been emphasised in previous studies (Padua et al., 2004; Schoenmakers, Uiterwaal, Gulmans, Gooskens, & Helders, 2005). Further, Olesen, Kiddoo, and Metcalfe (2013), found no association between shunted hydrocephalus and the PedsQL score. However, a literature review by Bakaniene et al. (2016) showed contradictory findings concerning the associations between levels of impairment and HRQoL in children with SB. Bakaniene et al. (2016) related these

contradictory findings to the heterogeneity in the study sample, study designs, determinants and outcome measures.

The quantitative data (Study III and IV) suggest that bladder and bowel dysfunction does not have a significant association with self-perceived health. This finding is both surprising and incongruent with the children's narratives in Study II, which revealed the impact of incontinence on the children's health and behaviours.

SOC showed a decline in the total score in Study IV. Moreover, the SOC scores reported by both the SB group and the reference group were low (47 ± 05 and 55 ± 14 , respectively) when compared with the cut-off points proposed by a previous study (Mendel et al., 2001), which considered a score of 35–60 to be low. Although this thesis does not measure the impact of sociocultural conditions, economic hardship and political violence on SOC, these factors might influence low SOC in the adolescents living in the West Bank, Palestine. Likewise, Pham, Vinck, Kinkodi, and Weinstein (2010) suggested that cumulative exposure to political violence is inversely associated with SOC among a general population in Eastern Democratic Republic of Congo. Studies of SOC among adolescents in different cultural groups have indicated that low SOC could be related to weak external GRR, low education, low socioeconomic status, different perceptions of the SOC construct and living in areas of political violence (Abu-Kaf, Braun-Lewensohn, & Kalagy, 2017; Braun-Lewensohn & Sagy, 2011).

To our knowledge, first-hand studies of SOC in adolescents with SB are lacking. Research has shown that people with a strong sense of coherence are more likely to identify stressful conditions, can handle stress better and are healthier than people with weak SOC (Lindmark, Hakeberg, & Hugoson, 2011; Super, Wagemakers, Picavet, Verkooijen, & Koelen, 2015). Further, Surtees et al. (2007) claimed that patients with a low SOC have less confidence in adapting to the difficulties faced in their lives than those with a higher SOC. Thus, the low SOC scores among adolescents with SB in Study IV might imply less ability to adjust to physical and social conditions.

Study IV found a positive association between the SOC and PedsQL scores, and these findings are consistent with a previous systematic review published by Eriksson and Lindström (2007) which noted the potential impact of SOC on perceived health. It was argued by Eriksson and Lindström (2007) that the stronger the SOC, the better the perceived health.

7 METHODOLOGICAL CONSIDERATIONS

Qualitative and quantitative approaches are used for complementary purposes to answer different research questions in nursing and other disciplines (Hammarberg, Kirkman, & De Lacey, 2016). The most critical issue in selecting a research design is its appropriateness to the aim (Polit & Beck, 2016). The methods used in this thesis were consistent with the aims presented in the four studies.

In qualitative studies, the purpose is to describe a phenomenon and create meaning and understanding by visualising the processes of people rather than outcomes (Suter, 2012). In this thesis, the qualitative studies contributed to health care science and demonstrated ways in which to care for vulnerable and marginalised children with SB and their families and how to meet their needs.

The key issue in qualitative studies is to generate in-depth data sufficient for illuminating the phenomenon under study; therefore, when selecting the sample and setting, it is important to consider information richness (Polit & Beck, 2016), as a small number of participants who are able to reflect on their experiences may generate a large amount of data for analysis. Further, sampling decisions in qualitative studies are not guided by the desire for generalisability to a target population. Instead, the principle guiding the collection of a population sample is to simply describe unique phenomena – in Study I and II, this meant that participants had to have experienced the actual phenomenon and were capable of expressing it, i.e., being the mother of a child with SB, and being a child with SB.

Conducting narrative interviews in Study I and II was beneficial, as the interviews generated sufficiently extensive data about mothers' and children's lived experiences. However, the knowledge acquired from these narratives would be of no value if it could not be spelled out and explained, as people's narratives often lack an explicit essence (Lindseth & Norberg, 2004). Therefore, the method chosen for the analysis was the phenomenological-hermeneutical method. This method, developed and described by Lindseth and Norberg (2004), was useful in fulfilling the aim of the study and in providing a deeper understanding of the complex multidimensional nature of the phenomenon. The method explored the participants' views, thoughts, perceptions and feelings about their lifeworld experiences.

The most important aspect that should be discussed in detail are the ethical issues introduced by studying children and, more sharply, when conducting

interviews with children, because their experiences and understandings of the world differ from those of adults and their manner of communication might also differ (Einarsdóttir, 2007). In Study II, III and IV, close attention was paid to the process of obtaining assent from the children and informed consent from their parents as well as clarifying the study objectives for them. Both children and parents were provided with understandable information about the interview questions in Study II to facilitate communication. In Study III and IV, detailed explanations were given to the two groups of children (children/adolescents with SB and healthy children/adolescents) and their parents to introduce them to the research topic.

However, several issues of trustworthiness must be considered and discussed in relation to qualitative studies. According to Lincoln and Guba (1985), trustworthiness includes four interrelated criteria: credibility dependability, conformability and transferability. Credibility concerns the richness of the data collected and confidence in the truth of the findings (Lincoln & Guba, 1985). To facilitate rich narratives from mothers and children (Study I and II), the participants were of different ages and lived in different areas; additionally, they were selected to exhibit wide variation in the level and type of deformities related to SB, thereby yielding variations in the data. All interviews started with one main question, but follow-up questions were used to encourage interviewees to elaborate on their narratives. The interpretation was based on participants' original narratives, and the interpretation process was described in detail. However, according to Ricoeur (1976), there is always more than one way to interpret a text, since texts convey many meanings, i.e., the phenomenological-hermeneutic approach does not offer one probable interpretation or truth.

Dependability is the criterion used to assess reliability (Hammarberg et al., 2016) and is closely related to credibility. It refers to whether the findings are consistent and can be repeated (Polit & Beck, 2016). Using the same interview guide for all the participants, including the same opening question and relatively similar follow-up questions, was expected to strengthen dependability. Conformability refers to the objectivity of the findings – that is, whether the findings reflect the participants' voices and thoughts or are in fact shaped more by the interviewer's preconceptions (Polit & Beck, 2016). In this regard, the researcher's preconceptions should be considered from the beginning of the analytical process through to the naïve understanding and structural analyses to the comprehensive understanding. However, the researcher's preconceptions can never be totally avoided and will as such always have some influence on the analysis. According to Dahlberg, Dahlberg, and Nystrom (2008), we cannot free ourselves from our preconceptions, but

we can be aware of them. This means that we can revise, broaden and deepen our preconceptions through critical reflection. According to Lindseth and Norberg (2004), preconceptions must be recognised, reflected upon and controlled during the analysis. I reflected on my preconceptions by writing them down, as presented in the introduction section.

The transferability of the findings in this thesis is best understood within paediatric and family nursing in the Palestinian health care context. Although the number of participants was small, the findings could be transferred to the context of mothers caring for children with SB and other disabilities, especially in the context of Arabic societies.

In quantitative studies, consideration must be given to the validity and reliability of the measures. The instruments used in this thesis (PedsQL and SOC) are well established, have been used extensively in the past, and are considered valid and reliable. Moreover, they have been validated in Arabic. The validity and reliability of the PedsQL™ 4.0 GCS, Arabic version, have been shown by Varni et al. (2005), while the SOC-13 Scale, Arabic version, has been shown to be valid and reliable by Eriksson and Lindström (2005). Although the PedsQL™ 4.0 has been used previously in children and adolescents with SB, the SOC has not been tested on this group. However, the results were related to SOC among children with other chronic illnesses.

Another important aspect in quantitative studies is the sampling process. In Studies III and IV, the researcher used convenience sampling, rather than probability sampling. In cases where information is limited, such as for children with SB in Palestine – particularly related to their self-perceived health and SOC – a convenience sample can be practical. Using this sampling method may help in describing such conditions; while convenience sampling creates limitations with respect to the generalizability of results (Etikan, Musa, & Alkassim, 2016) it was sufficient to meet the purposes of the current studies, which were cross-sectional and descriptive, with few attempts to verify relationships between the variables.

However, one must consider that neither the descriptive information nor the relationships found between the variables in Studies III and IV should be interpreted as applicable to the full range of children and adolescents with SB in other contexts. At best, the studies may be representative of the segment of such children who are living in Palestine. Caution must be taken in generalising descriptive information, or making inferences, due to the lack of probability sampling. However, the use of a purposive sample and the small number of participants in the qualitative studies was not problematic, because

generalising to a population is not the purpose of such research. Instead, the purpose of the qualitative study in this area was to understand the lived experiences of the mothers and children, as they gave rich and detailed descriptions of their experiences.

This thesis has some limitations related to the sample that should be considered. Children with intellectual deficits were excluded, yet these children's experiences with vulnerability and social exclusion may be different from those of the children interviewed in this study. The study relied on the voices of children registered in governmental and NGO organisations. Children who are particularly marginalised or hidden due to negative sociocultural attitudes and the stigma of disability, as well as those who do not have accessibility to services, were not considered eligible. Further, the quantitative studies were cross-sectional, and the effects of SB on self-perceived health and SOC could not be established from this study.

8 CONCLUSION AND IMPLICATIONS

To the best of our knowledge, this is the first thesis in the context of the West Bank, Palestine, that has used qualitative and quantitative data to contribute first-hand knowledge about the lived experiences, self-perceived health and SOC of children and adolescents with SB.

The findings in Studies I and II showed that the participants experienced suffering related to SB consequences, and found their life situation burdensome. Both the mothers and children struggled with negative social attitudes and prejudice that led to social exclusion. The mothers found, lack of knowledge about SB, daily stressors and challenges of caring for the sick child, overwhelming. Despite the stressful life conditions, the mothers showed adjustment abilities. The children had a negative self-concept, mainly related to their impairments, and particularly the difficulties they faced with incontinence problems and the use of CIC. They showed warning behavioral problems and feelings of shame, secrecy, and abandonment.

In Studies III and IV, low levels of self-perceived health and SOC among children and adolescents with SB, in comparison to their healthy peers, were found. Social functioning was scored low among adolescents with SB, but was lowest in children. The findings showed that mobility impairment and shunted hydrocephaly were inversely associated with self-perceived health. Further, SOC was positively associated with self-perceived health among adolescents.

The thesis shows how the complex consequences of SB and the contextual circumstances in Palestine – including stigma, poverty and ongoing political conflict – affect the mothers' and children's lives. These factors emerged as important challenges to guide paediatric nurses, rehabilitation nurses and other health care providers in planning child and family care. In particular, strengthening the role of the family by enhancing and mobilising their inherent supportive capabilities, while simultaneously reducing exposure to risk experiences, is a central part of facilitating positive psychosocial adjustment and adaptation to living with SB in Palestine.

Major findings from this thesis suggest that

- Nurses, and especially paediatric nurses, need to have better knowledge about SB and what nursing actions can be helpful for children with SB and their families.

- Nurses should take an active role in controlling bladder and bowel incontinence, by teaching mothers about the use of CIC and other helpful remedies for controlling bowel movements, and teaching children at an early age how to use CIC and other caring remedies.
- Nurses can play an important role in enhancing children's activities by teaching them and their families about the importance of physical activities and commitment to the exercises schedule that supports their ambulation levels.
- The mothers' roles in the care of children with SB and their efforts to maintain their family's well-being call for more coordinated health care interventions for families (i.e., FCC policies and CBR programs), which would both enable and empower the children and their families.
- Adoption of and adherence to the FCC continuum within the provision of health services in Palestine would be expected to help Palestinian children with SB and their families to face the stigma of the illness, thereby increasing their participation, independence and societal acceptance.
- It is suggested that the PedsQL™ 4.0 and SOC-13 measures to be used continually by nurses in rehabilitation centres and routinely to assess any changes in the children's physical and psychosocial health and SOC.
- As social functioning received low scores on the PedsQL subscales, social support services for children and adolescents with SB and other disabilities are sorely needed in Palestine to promote these children's social functioning, autonomy and independence.
- Knowledge generated from this thesis could help nurses, health professionals, families and stakeholders to collaborate in the establishment of health and social network services. These efforts could be used to further promote CBR services and develop national guidelines of care for children with SB and their families in Palestine.

9 FUTURE RESEARCH

Spina bifida is a burden that affects the lives of children, their mothers and their families. As this was the first study in the West Bank, Palestine, further studies are needed to provide a basis for health care policies to meet the growing needs of children with SB and their families, and to support future health care strategies for this group of children.

- Given the limited data available on SB in Palestine, epidemiological studies are recommended to explore its incidence, prevalence, life expectancy and complications.
- This thesis found that social attitudes have the greatest impact on mothers' and children's psychosocial health. Future studies are needed to explore the knowledge, attitudes, beliefs and behaviours of health care providers about SB in Palestine. Further, to obtain their knowledge about CBR services provided for children with SB.
- Research should seek to understand the role of nurses and other health care providers in supporting this group of patients and their families. Emphasis should be placed on assessing the extent of their interactions and collaboration with families.
- Intervention studies are needed that would conduct the FCC in one of the rehabilitation centres to determine whether the FCC strategies are applicable to Palestinian society in various settings and could create benefits for families.
- An important recommendation for future studies is to assess the knowledge of children and their families about urinary incontinence and its impact on children's health.
- Given the supportive role of husbands and other family members, further interview studies are recommended that would involve fathers and siblings of children with SB in order to understand their experiences.

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APPENDIX

Interview guide Study I

- ✓ Can you tell me about your experience: How it is to be a mother to a child with Spina Bifida? Follow-up questions like -Can you tell me more about -. How did you feel when? What do you mean? Can you give an example, how it felt, and so on.
- ✓ Can you describe your experience and your feelings in the first time when you understand that you would have a baby with SB? Follow-up questions Like- who told you about it. How you react to this? What you think about it? Can you tell me more about this?
- ✓ Can you describe your experience and expectations about your baby before delivery? Tell me about your experience when your child was born with Spina Bifida at the delivery time and after delivery.
- ✓ Can you tell me about the reaction of people around you when they heard about it and when they saw the baby? How does this, to have a child with SB affects your family life? What are the most difficulties you faced since the delivery of your child- How do you feel now with the number of years - what you hope?
- ✓ Can you tell me about your experience with the child's treatment? How did it go- How do you feel about it.
- ✓ Please tell me about your feelings after the number of year passed with the child what do you think how you experience the life and the future life

Interview guide Study II

- ✓ Can you tell me about your daily life experience with SB?
- ✓ Can you tell me about a typical day? What do you do every day?
- ✓ Can you describe your experience with your family in the home environment?
- ✓ Tell me about one day you spend with your friends. How do you feel about it?
- ✓ Can you tell me about one day you spent in the hospital?
- ✓ What about the equipment and devices you of the use?

REFERENCES

- Abdul-Rasoul, M., AlOtaibi, F., AlMahdi, M., & AlKandari, H. (2012). Reliability and validity of the Arabic version of the PedsQLTM 4.0 generic ore scales and PedsQLTM 3.0 diabetes module. *Journal of Diabetes Mellitus*, 2(03), 301. <http://dx.doi.org/10.4236/jdm.2012.23047>.
- Abresch, R. T., McDonald, D. A., Widman, L. M., McGinnis, K., & Hickey, K. J. (2007). Impact of spinal cord dysfunction and obesity on the health-related quality of life of children and adolescents. *The journal of spinal cord medicine*, 30(sup1), S112-S118. <https://www.ncbi.nlm.nih.gov/pmc/articles/PMC2031984/>.
- Abu-Kaf, S., Braun-Lewensohn, O., & Kalagy, T. (2017). Youth in the midst of escalated political violence: sense of coherence and hope among Jewish and Bedouin Arab adolescents. *Child Adolesc Psychiatry Ment Health*, 11(1), 42.
- Antle, B. J., Montgomery, G., & Stapleford, C. (2009). The Many Layers of Social Support: Capturing the Voices of Young People with Spina Bifida and Their Parents. *Health & Social Work*, 34(2), 97-106.
- Antonovsky, A. (1979). *Health, stress, and coping* (Vol. 1.). San Francisco: Jossey-Bass
- Arango, P. (2011). Family-centered care. *Academic Pediatrics*, 11(2), 97-99.
- Assembly, U. G. (1948). Universal declaration of human rights. *UN General Assembly*.
- Au, K. S., Ashley Koch, A., & Northrup, H. (2010). Epidemiologic and genetic aspects of spina bifida and other neural tube defects. *Developmental disabilities research reviews*, 16(1), 6-15. <http://dx.doi.org/10.1002/ddrr.93>.
- Bakaniene, I., Prasauskiene, A., & Vaiciene Magistris, N. (2016). Health-related quality of life in children with myelomeningocele: a systematic review of the literature. *Child: Care, Health and Development*, 42(5), 625-643. <http://dx.doi.org/10.1111/cch.12367>.
- Bannink, F., Idro, R., & van Hove, G. (2016). Parental stress and support of parents of children with spina bifida in Uganda.(Original Research). *African Journal of Disability*, 5(1). <http://dx.doi.org/10.4102/ajod.v5i1.225>.
- Barakat, H. (1993). *The Arab world: Society, culture, and state* (pp. 7-48). Retrieved from http://acc.teachmideast.org/texts.php?module_id=8&reading_id=13
- Barbosa, M. A. M., Balieiro, M. M. F. G., & Pettengill, M. A. M. (2012). Family-centered care in the context of children with disabilities and

- their families: a reflective review. *Texto & Contexto-Enfermagem*, 21(1), 194-199.
- Beauchamp, T. L., & Childress, J. F. (2008). *Principles of biomedical ethics*: Oxford University Press, USA
- Behnke, E. A. (2011). Husserl's phenomenology of embodiment.
- Blake, F. G. (1954). *The child, his parents and the nurse*: Lippincott
- Bratt, E.-L., & Moons, P. (2015). Forty years of quality-of-life research in congenital heart disease: Temporal trends in conceptual and methodological rigor. *International journal of cardiology*, 195, 1-6. <http://dx.doi.org/10.1016/j.ijcard.2015.05.070>.
- Braun-Lewensohn, O., & Sagy, S. (2011). Salutogenesis and culture: Personal and community sense of coherence among adolescents belonging to three different cultural groups: Taylor & Francis.
- Brekke, I., Früh, E. A., Kvarme, L. G., & Holmstrøm, H. (2017). Long-time sickness absence among parents of pre-school children with cerebral palsy, spina bifida and down syndrome: a longitudinal study. *BMC Pediatrics*, 17(1), 26.
- Burke, R., & Liptak, G. S. (2011). Providing a primary care medical home for children and youth with spina bifida. *Pediatrics*, 128(6), e1645-e1657. doi:10.1542/peds.2011-2219
- Burton, G., Sayrafi, I., & Srour, S. A. (2013). Inclusion or transformation? An early assessment of an empowerment project for disabled people in occupied Palestine. *Disability & Society*, 28(6), 812-825. <http://dx.doi.org/10.1080/09687599.2013.802223>.
- Cocks, A. (2008). Researching the lives of disabled children: the process of participant observation in seeking inclusivity. *Qualitative Social Work*, 7(2), 163-180. <http://dx.doi.org/10.1177/1473325008089628>.
- Connors, C., & Stalker, K. (2007). Children's experiences of disability: pointers to a social model of childhood disability. *Disability & Society*, 22(1), 19-33. <http://dx.doi.org/10.1080/09687590601056162>.
- Copp, A. J., Adzick, N. S., Chitty, L. S., Fletcher, J. M., Holmbeck, G. N., & Shaw, G. M. (2015). Spina bifida. *Nature reviews. Disease primers* 1, 15007. <http://dx.doi.org/10.1038/nrdp.2015.7>.
- Coughlin, M. B., & Sethares, K. A. (2017). Chronic sorrow in parents of children with a chronic illness or disability: an integrative literature review. *Journal of Pediatric Nursing*, 37, 108-116.
- Crabtree, S. A. (2007). Family responses to the social inclusion of children with developmental disabilities in the United Arab Emirates. *Disability & Society*, 22(1), 49-62.
- Dahlberg, K., Dahlberg, H., & Nystrom, M. (2008). *Reflective lifeworld research*: Studentlitteratur, Lund

- David, R. (2008). The care of a person with a diseased and broken heart. *Journal of Pastoral Care & Counseling*, 62(4), 363-366.
- Dennis, M., & Barnes, M. A. (2010). The cognitive phenotype of spina bifida meningocele. *Developmental disabilities research reviews*, 16(1), 31-39. <http://dx.doi.org/10.1002/ddrr.89>.
- Dicianno, B. E., Kurowski, B. G., Yang, J. M. J., Chancellor, M. B., Bejjani, G. K., Fairman, A. D., . . . Sotirake, J. (2008). Rehabilitation and medical management of the adult with spina bifida. *American journal of physical medicine & rehabilitation*, 87(12), 1027-1050. <http://dx.doi.org/10.1097/PHM.0b013e31818de070>.
- Dunleavy, M. J. (2007). The role of the nurse coordinator in spina bifida clinics. *The Scientific World Journal*, 7, 1884-1889.
- Eide, A. H. (2006). Impact of Community-Based Rehabilitation Programmes: The Case of Palestine. *Scandinavian Journal of Disability Research*, 8(4), 199-210. doi:<http://dx.doi.org/10.1080/15017410500466750>
- Einarsdóttir, J. (2007). Research with children: Methodological and ethical challenges. *European early childhood education research journal*, 15(2), 197-211. <https://doi.org/10.1080/13502930701321477>.
- Ekman, I., Swedberg, K., Taft, C., Lindseth, A., Norberg, A., Brink, E., . . . Sunnerhagen, K. S. (2011). Person-Centered Care — Ready for Prime Time. *European Journal of Cardiovascular Nursing*, 10(4), 248-251. <http://dx.doi.org/10.1016/j.ejcnurse.2011.06.008>.
- Elkinton, J. R. (1966). Medicine and the quality of life. *Annals of Internal Medicine*, 64(3), 711-714.
- Eriksson, M., & Lindström, B. (2005). Validity of Antonovsky's sense of coherence scale: a systematic review. *Journal of Epidemiology & Community Health*, 59(6), 460-466.
- Eriksson, M., & Lindström, B. (2007). Antonovsky's sense of coherence scale and its relation with quality of life: a systematic review. *Journal of Epidemiology & Community Health*, 61(11), 938-944.
- Eriksson, M., & Lindström, B. (2008). A salutogenic interpretation of the Ottawa Charter. *Health promotion international*, 23(2), 190-199.
- Eriksson, M., Lindström, B., & Lilja, J. (2007). A sense of coherence and health. Salutogenesis in a societal context: Åland, a special case? *Journal of epidemiology and community health*, 61(8), 684-688.
- Etikan, I., Musa, S. A., & Alkassim, R. S. (2016). Comparison of convenience sampling and purposive sampling. *American Journal of Theoretical and Applied Statistics*, 5(1), 1-4.
- Fayers, P. M., & Machin, D. (2013). *Quality of life: the assessment, analysis and interpretation of patient-reported outcomes*. Chichester: John Wiley & Sons
- Finlay, L. (2012). Debating phenomenological methods *Hermeneutic phenomenology in education* (pp. 17-37). SensePublishers: Springer.

- Fischer, N., Church, P., Lyons, J., & McPherson, A. C. (2015). A qualitative exploration of the experiences of children with spina bifida and their parents around incontinence and social participation. *Child: Care, Health and Development*, 41(6), 954-962. <http://dx.doi.org/10.1111/cch.12257>.
- Fonseca, E. B. d., Raskin, S., & Zugaib, M. (2013). Folic acid for the prevention of neural tube defects. *Revista Brasileira de Ginecologia e Obstetrícia*, 35(7), 287-289. <http://dx.doi.org/doi10.1590/S0100-72032013000700001>.
- Gazarian, P. K., Henneman, E. A., & Chandler, G. E. (2010). Nurse decision making in the prearrest period. *Clinical Nursing Research*, 19(1), 21-37.
- Giacaman, R. (2001). A community of citizens: disability rehabilitation in the Palestinian transition to statehood. *Disability & Rehabilitation*, 2001, Vol.23(14), p.639-644, 23(14), 639-644. doi:10.1080/09638280110036544
- Giacaman, R., Khatib, R., Shabaneh, L., Ramlawi, A., Sabri, B., Sabatinelli, G., . . . Laurance, T. (2009). Health status and health services in the occupied Palestinian territory. *Lancet*, 373(9666), 837-849. [http://dx.doi.org/10.1016/S0140-6736\(09\)60107-0](http://dx.doi.org/10.1016/S0140-6736(09)60107-0).
- Giacaman, R., Rabaia, Y., Nguyen-Gillham, V., Batniji, R., Punamäki, R.-L., & Summerfield, D. (2011). Mental health, social distress and political oppression: The case of the occupied Palestinian territory. *Global Public Health*, 6(5), 547-559. <http://dx.doi.org/10.1080/17441692.2010.528443>.
- Goffman, E. (1963). Stigma: Notes on a spoiled identity. *New York: Simon & Shuster*.
- Greenley, R. N. (2010). Health professional expectations for self-care skill development in youth with spina bifida. *Pediatric nursing*, 36(2), 98. http://theshinelibrary.org/content/publications%20_documents/Jou087.pdf.
- Hammarberg, K., Kirkman, M., & De Lacey, S. (2016). Qualitative research methods: when to use them and how to judge them. *Human Reproduction*, 31(3), 498-501. <https://doi.org/10.1093/humrep/dev334>.
- Hanan, F. A. R., Wick, L., Halileh, S., Hassan-Bitar, S., Chekir, H., Watt, G., & Khawaja, M. (2009). Health in the Occupied Palestinian Territory 2: Maternal and child health in the occupied Palestinian territory. *The Lancet*, 373(9667), 967-977. [https://doi.org/10.1016/S0140-6736\(09\)60108-2](https://doi.org/10.1016/S0140-6736(09)60108-2). doi:10.1016/S0140-6736(09)60108-2

- Harrison, T. M. (2010). Family-Centered Pediatric Nursing Care: State of the Science. *Journal of Pediatric Nursing*, 25(5), 335-343.
<https://doi.org/10.1016/j.pedn.2009.01.006>.
- Hasnain, R., Shaikh, L. C., & Shanawani, H. (2008). Disability and the Muslim perspective: An introduction for rehabilitation and health care providers. Center for International Rehabilitation Research Information and Exchange, University at Buffalo, State University of New York. <http://cirrie.buffalo.edu/culture/monographs/muslim.pdf>.
- Helseth, S., & Misvær, N. (2010). Adolescents' perceptions of quality of life: what it is and what matters. *Journal of Clinical Nursing*, 19(9-10), 1454-1461.
- Hemingway, A. (2011). Lifeworld-led care: Is it relevant for well-being and the fifth wave of public health action? *International Journal of Qualitative Studies on health and Well-being*, 6(4), 10364.
- Hockenberry, M. J., & Wilson, D. (2003). *Wong's nursing care of infants and children* Elsevier Health Sciences 146-148
- Hockenberry, M. J., & Wilson, D. (2018). *Wong's nursing care of infants and children* Elsevier Health Sciences 1312-1318
- Hollway, W., & Jefferson, T. (2008). The free association narrative interview method.
- Holmbeck, G. N., & Devine, K. A. (2010). Psychosocial and family functioning in spina bifida. *Developmental disabilities research reviews*, 16(1), 40-46.
- Holmbeck, G. N., Westhoven, V. C., Phillips, W. S., Bowers, R., Gruse, C., Nikolopoulos, T., . . . Davison, K. (2003). A Multimethod, Multi-Informant, and Multidimensional Perspective on Psychosocial Adjustment in Preadolescents With Spina Bifida. *Journal of Consulting and Clinical Psychology*, 71(4), 782-796.
doi:10.1037/0022-006X.71.4.782
- Husserl, E. (1970). *The crisis of European sciences and transcendental phenomenology: An introduction to phenomenological philosophy*: Northwestern University Press
- IBM SPSS Statistics 24. (2016). IBM Corporation. SPSS for Windows, Version 24.
- Isaac, J., & Hilal, J. (2011). Palestinian landscape and the Israeli–Palestinian conflict. *International Journal of Environmental Studies*, 68(4), 413-429. <http://dx.doi.org/10.1080/00207233.2011.582700>.
- Jahoda, A., Wilson, A., Stalker, K., & Cairney, A. (2010). Living with stigma and the self-perceptions of people with mild intellectual disabilities. *Journal of Social Issues*, 66(3), 521-534.
<http://dx.doi.org/10.1111/j.1540-4560.2010.01660.x>.

- Jarar, A. (2009). Disability in Palestine: realities and perspectives. *This Week in Palestine*, 137.
<http://archive.thisweekinpalestine.com/details.php?id=2868>.
- Jenkinson, M. D., Campbell, S., Hayhurst, C., Clark, S., Kandasamy, J., Lee, M. K., . . . Mallucci, C. L. (2011). Cognitive and functional outcome in spina bifida-Chiari II malformation. *Childs Nerv Syst*, 27(6), 967-974. <http://dx.doi.org/10.1007/s00381-010-1368-7>.
- Johansen, H., Dammann, B., Andresen, I.-L., & Fagerland, M. W. (2013). Health-related quality of life for children with rare diagnoses, their parents' satisfaction with life and the association between the two. *Health and quality of life outcomes*, 11(1), 1.
[doi:https://hqlo.biomedcentral.com/articles/10.1186/1477-7525-11-152](https://hqlo.biomedcentral.com/articles/10.1186/1477-7525-11-152)
- Johnson, E., Nilsson, S., & Adolfsson, M. (2015). Eina! Ouch! Eish! Professionals' Perceptions of How Children with Cerebral Palsy Communicate About Pain in South African School Settings: Implications for the use of AAC. *Augment Altern Commun*, 31(4), 325-335. <http://www.ncbi.nlm.nih.gov/pubmed/26372118>.
[doi:10.3109/07434618.2015.1084042](https://doi.org/10.3109/07434618.2015.1084042)
- Jones, E. E. (1984). *Social stigma: The psychology of marked relationships*: WH Freeman
- Kamenetsky, S. B., Dimakos, C., Aslemand, A., Saleh, A., & Ali-Mohammed, S. (2016). Eliciting help without pity: the effect of changing media images on perceptions of disability. *Journal of social work in disability & rehabilitation*, 15(1), 1-21.
<http://dx.doi.org/10.1080/1536710X.2016.1124251>.
- Karimi, M., & Brazier, J. (2016). Health, Health-Related Quality of Life, and Quality of Life: What is the Difference? *PharmacoEconomics*, 1-5.
<https://doi.org/10.1007/s40273-016-0389-9>.
- Kaur, Iqbal, Hillis, Samira Ahmed, Abu Al-Ghaib, Ola, & . . . Montserrat; Essama, S. (2016). West Bank and Gaza - Disability in the Palestinian territories: assessing situation and services for people with disabilities (English). *Washington, D.C. : World Bank Group*.
<http://documents.worldbank.org/curated/en/501421472239948627/West-Bank-and-Gaza-Disability-in-the-Palestinian-territories-assessing-situation-and-services-for-people-with-disabilities>.
- Khamis, V. (2008). Post-traumatic stress and psychiatric disorders in Palestinian adolescents following intifada-related injuries. *Social Science & Medicine*, 67(8), 1199-1207.
<http://dx.doi.org/doi:10.1016/j.socscimed.2008.06.013>.
[doi:http://dx.doi.org/](http://dx.doi.org/)
- Lang, R., Kett, M., Groce, N., & Trani, J.-F. (2011). Implementing the United Nations Convention on the rights of persons with disabilities:

- principles, implications, practice and limitations. *ALTER-European Journal of Disability Research/Revue Européenne de Recherche sur le Handicap*, 5(3), 206-220.
<http://dx.doi.org/10.1016/j.alter.2011.02.004>.
- Leonardi, M., Bickenbach, J., Ustun, T. B., Kostanjsek, N., & Chatterji, S. (2006). The definition of disability: what is in a name? *The Lancet*, 368(9543), 1219-1221. [https://doi.org/10.1016/S0140-6736\(06\)69498-1](https://doi.org/10.1016/S0140-6736(06)69498-1).
- Lin, X.-J., Lin, I.-M., & Fan, S.-Y. (2013). Methodological issues in measuring health-related quality of life. *Tzu Chi Medical Journal*, 25(1), 8-12. <https://doi.org/10.1016/j.tcmj.2012.09.002>.
- Lincoln, Y. S., & Guba, E. G. (1985). *Naturalistic inquiry*. Beverly Hills, Calif: Sage
- Lindmark, U., Hakeberg, M., & Hugoson, A. (2011). Sense of coherence and its relationship with oral health-related behaviour and knowledge of and attitudes towards oral health. *Community dentistry and oral epidemiology*, 39(6), 542-553.
- Lindsay, S. (2014). A Qualitative Synthesis of Adolescents' Experiences of Living With Spina Bifida. *Qualitative Health Research*, 24(9), 1298-1309. <http://dx.doi.org/10.1177/1049732314546558>.
- Lindseth, A., & Norberg, A. (2004). A phenomenological hermeneutical method for researching lived experience. *Scand J Caring Sci*, 18(2), 145-153. <http://dx.doi.org/10.1111/j.1471-6712.2004.00258.x>.
- Liptak, G. S., Garver, K., & Dosa, N. P. (2013). Spina bifida grown up. *Journal of developmental and behavioral pediatrics : JDBP*, 34(3), 206-215. <http://dx.doi.org/10.1097/DBP.0b013e31828c5f88>.
- MacGregor, D. G., & Godfrey, J. (2011). Arab cultural influences on intertemporal reasoning. *Available at SSRN 1966639*. Retrieved from http://papers.ssrn.com/sol3/papers.cfm?abstract_id=1966639.
- Major, B., & O'Brien, L. T. (2005). The social psychology of stigma. *Annu. Rev. Psychol.*, 56, 393-421.
- Marie, M., Hannigan, B., & Jones, A. (2016). Mental health needs and services in the West Bank, Palestine. *International journal of mental health systems*, 10(1), 23.
- Mataria, A., Khatib, R., Donaldson, C., Bossert, T., Hunter, D. J., Alsayed, F., & Moatti, J.-P. (2009). The health-care system: an assessment and reform agenda. *The Lancet*, 373(9670), 1207-1217.
- Mendel, B., Bergenius, J., & Langius, A. (2001). The sense of coherence: A tool for evaluating patients with peripheral vestibular disorders. *Clinical Otolaryngology and Allied Sciences*, 26(1), 19-24.
<http://dx.doi.org/10.1046/j.1365-2273.2001.00398.x>.
- Merleau-Ponty, M. (2004). *The world of perception*: Routledge. Taylor&francis group. London and Newyork.

- Monsen, R. B. (1999). Mothers' experiences of living worried when parenting children with spina bifida. *Journal of Pediatric Nursing*, 14(3), 157-163.
- Müller Godeffroy, E., Michael, T., Poster, M., Seidel, U., Schwarke, D., & Thyen, U. (2008). Self-reported health-related quality of life in children and adolescents with myelomeningocele. *Developmental Medicine & Child Neurology*, 50(6), 456-461.
<https://onlinelibrary.wiley.com/doi/abs/10.1111/j.1469-8749.2008.02054.x>.
- Muylaert, C. J., Sarubbi Jr, V., Gallo, P. R., Neto, M. L. R., & Reis, A. O. A. (2014). Narrative interviews: an important resource in qualitative research. *Revista da Escola de Enfermagem da USP*, 48(SPE2), 184-189.
- Naghi, J., Philip, K., Phan, A., Cleenewerck, L., & Schwarz, E. (2012). The Effects of Spirituality and Religion on Outcomes in Patients with Chronic Heart Failure (Vol. 51, pp. 1124-1136). Boston.
- Nahal, M., Axelsson, A. B., Imam, A., & Wigert, H. (2019). Palestinian children's narratives about living with spina bifida: Stigma, vulnerability, and social exclusion. *Child: Care, Health and Development*, 45(1), 54. doi:10.1111/cch.12625
- Nilsson , & Qutteina, M. (2005). Evaluation of the CBR program in Palestine—from the perspective of persons with disabilities themselves. *Ramallah: Diakonia and NAD*.
- Nilsson, K. W., Leppert, J., Simonsson, B., Starrin, B., Karlstads, u., Avdelningen för sociala, s., & Fakulteten för samhälls- och, l. (2010). Sense of coherence and psychological well-being: improvement with age. *Journal of Epidemiology and Community Health* (1979-), 64(4), 347-352. <http://dx.doi.org/10.1136/jech.2008.081174>.
- Olesen, J. D., Kiddoo, D. A., & Metcalfe, P. D. (2013). The association between urinary continence and quality of life in paediatric patients with spina bifida and tethered cord. *Paediatrics & child health*, 18(7), e32-e38. <https://doi-org.ezproxy.ub.gu.se/10.1093/pch/18.7.e32>.
- Olsson, M., Hansson, K., Lundblad, A. M., & Cederblad, M. (2006). Sense of coherence: definition and explanation. *International Journal of Social Welfare*, 15(3), 219-229. doi:10.1111/j.1468-2397.2006.00410.x
- Ong, L. C., Norshireen, N. A., & Chandran, V. (2011). A comparison of parenting stress between mothers of children with spina bifida and able-bodied controls. *Developmental Neurorehabilitation*, 14(1), 22-28. <http://dx.doi.org/10.3109/17518423.2010.523057>.
- Padua, L., Rendeli, C., Ausili, E., Aprile, I., Caliandro, P., Tonalì, P., & Salvaggio, E. (2004). Relationship between the clinical-

- neurophysiologic pattern, disability, and quality of life in adolescents with spina bifida. *Journal of child neurology*, 19(12), 952-957.
- Palestinian Central Bureau of Statistics. (2016). *Palestine in figures 2015*. Ramallah-Palestine Retrieved from <http://www.pcbs.gov.ps/Downloads/book2115.pdf>.
- Palestinian Central Bureau of Statistics & Ministry of Social Affairs. (2011). *Disability survey 2011: Press conference report*. Ramallah: Palestine. Retrieved from http://www.washingtongroup-disability.com/wp-content/uploads/2016/03/disability_e2011.pdf.
- Peny-Dahlstrand, M., Krumlinde-Sundholm, L., & Gosman-Hedström, G. (2012). Is autonomy related to the quality of performance of everyday activities in children with spina bifida? *Disability and Rehabilitation*, 34(6), 514-521. doi:10.3109/09638288.2011.610495
- Pham, P. N., Vinck, P., Kinkodi, D. K., & Weinstein, H. M. (2010). Sense of coherence and its association with exposure to traumatic events, posttraumatic stress disorder, and depression in eastern Democratic Republic of Congo. *Journal of traumatic stress*, 23(3), 313-321.
- Polit, D. F., & Beck, C. T. (2016). *Nursing research: Generating and assessing evidence for nursing practice* (Vol. 10th edition). Philadelphia: Philadelphia: Wolters Kluwer.Lippincott Williams & Wilkins
- Psihogios, A. M., Kolbuck, V., & Holmbeck, G. N. (2015). Condition self-management in pediatric spina bifida: A longitudinal investigation of medical adherence, responsibility-sharing, and independence skills. *J Pediatr Psychol*, 40(8), 790-803. <https://doi.org/10.1093/jpepsy/jsv044>.
- Ramachandra, P., Palazzi, K. L., Skalsky, A. J., Marietti, S., & Chiang, G. (2013). Shunted hydrocephalus has a significant impact on quality of life in children with spina bifida. *PM&R*, 5(10), 825-831. <https://doi.org/10.1016/j.pmrj.2013.05.011>.
- Richardson, B. (2010). The Disability Discrimination Act 1995. *Nursing And Residential Care*, 12(2), 93-97. <http://dx.doi.org/10.12968/nrec.2010.12.2.46113>.
- Ricoeur, P. (1976). *Interpretation theory: Discourse and the surplus of meaning*. FortWorth, TX: Christian University Press.
- Ricoeur, P. (1981). *Hermeneutics and the human sciences: Essays on language, action and interpretation*: Cambridge university press
- Rofail, D., Maguire, L., Heelis, R., Colligs, A., Lindemann, M., & Abetz, L. (2012). The impact of spina bifida on caregivers. *Neurol Ther*, 1(1), 4. <http://www.ncbi.nlm.nih.gov/pubmed/26000210>. doi:10.1007/s40120-012-0004-8
- Ryan, C. (2015). Everyday resilience as resistance: Palestinian women practicing sumud. *International Political Sociology*, 9(4), 299-315.

- Sawin, K. J., Bellin, M. H., Roux, G., Buran, C. F., & Brei, T. J. (2009). The experience of self-management in adolescent women with spina bifida. *Rehabil Nurs*, 34(1), 26-38. doi:10.1002/j.2048-7940.2009.tb00245.x
- Sawyer, S. M., & Macnee, S. (2010). Transition to adult health care for adolescents with spina bifida: research issues. *Developmental disabilities research reviews*, 16(1), 60-65. <http://dx.doi.org/10.1002/ddrr.98>.
- Schoenmakers, M., Uiterwaal, C., Gultmans, V. A. M., Gooskens, R., & Helders, P. J. M. (2005). Determinants of functional independence and quality of life in children with spina bifida. *Clinical Rehabilitation*, 19(6), 677-685.
- Shakespeare, T., & Watson, N. (2001). The social model of disability: an outdated ideology? *Exploring theories and expanding methodologies: Where we are and where we need to go* (pp. 9-28): Emerald Group Publishing Limited.
- Shields, N., Taylor, N. F., & Dodd, K. J. (2008). Self-concept in children with spina bifida compared with typically developing children. *Developmental Medicine and Child Neurology*, 50(10), 733-743. <http://dx.doi.org/10.1111/j.1469-8749.2008.03096.x>.
- Stuber, J., Meyer, I., & Link, B. (2008). Stigma, prejudice, discrimination and health. *Social Science & Medicine*, 67(3), 351-357. <http://dx.doi.org/10.1016/j.socscimed.2008.03.023>.
- Super, S., Wagemakers, M., Picavet, H., Verkooijen, K., & Koelen, M. (2015). Strengthening sense of coherence: opportunities for theory building in health promotion. *Health promotion international*, 31(4), 869-878.
- Surtees, P. G., Wainwright, N. W., Luben, R. L., Wareham, N. J., Bingham, S. A., & Khaw, K.-T. (2007). Adaptation to social adversity is associated with stroke incidence: evidence from the EPIC-Norfolk prospective cohort study. *Stroke*, 38(5), 1447-1453.
- Suter, W. N. (2012). Qualitative data, analysis, and design. *Introduction to educational research: A critical thinking approach*, 2, 342-386.
- Tennant, P. W., Pearce, M. S., Bythell, M., & Rankin, J. (2010). 20-year survival of children born with congenital anomalies: a population-based study. *The Lancet*, 375(9715), 649-656. [http://dx.doi.org/10.1016/S0140-6736\(09\)61922-X](http://dx.doi.org/10.1016/S0140-6736(09)61922-X).
- Thomas, C. (2010). Medical sociology and disability theory *New directions in the sociology of chronic and disabling conditions* (pp. 37-56): Springer.
- Todres, L., Galvin, K., & Dahlberg, K. (2007). Lifeworld-led healthcare: revisiting a humanising philosophy that integrates emerging trends.

- Medicine, health care and philosophy*, 10(1), 53-63.
<http://dx.doi.org/10.1007/s11019-006-9012-8>.
- Trudell, A. S., & Odibo, A. O. (2014). Diagnosis of spina bifida on ultrasound: always termination? *Best practice & research. Clinical obstetrics & gynaecology*, 28(3), 367-377.
<http://dx.doi.org/10.1016/j.bpobgyn.2013.10.006>.
- UNCRPD. (2006). Convention on the Rights of Persons with Disabilities
G.A. Res. 61/106, U.N. Doc. A/RES/61/1106.
http://www.un.org/en/development/desa/population/migration/generalassembly/docs/globalcompact/A_RES_61_106.pdf.
- Unicef. (1989). Convention on the Rights of the Child.
- UNICEF. (2016). Every child counts: understanding the needs and perspectives of children with disabilities in the State of Palestine.
http://www.unicef.org/oPt/ODI_Report_01-06-2017_FINAL.pdf.
- Vande Velde, S., Laridaen, J., Van Hoecke, E., Van Biervliet, S., De Bruyne, R., Van Winckel, M., & Goubert, L. (2016). Development and validation of a spina bifida-specific pediatric quality of life questionnaire: the Spina Bifida Pediatric Questionnaire, SBPQ. *Childs Nerv Syst*, 32(1), 105-110.
<http://www.ncbi.nlm.nih.gov/pubmed/26362680>.
doi:10.1007/s00381-015-2903-3
- Varni, J. W., Burwinkle, T. M., & Seid, M. (2005). The PedsQL™ as a pediatric patient-reported outcome: Reliability and validity of the PedsQL™ Measurement Model in 25,000 children. *Expert review of pharmacoeconomics & outcomes research*, 5(6), 705-719.
<http://dx.doi.org/10.1586/14737167.5.6.705>.
- Vermaes, I. P., Janssens, J. M., Mullaart, R. A., Vinck, A., & Gerris, J. R. (2008). Parents' personality and parenting stress in families of children with spina bifida. *Child: Care, Health and Development*, 34(5), 665-674. <http://dx.doi.org/10.1111/j.1365-2214.2008.00868.x>
- Vinck, A., Nijhuis-van der Sanden, M. W., Roeleveld, N. J., Mullaart, R. A., Rotteveel, J. J., & Maassen, B. A. (2010). Motor profile and cognitive functioning in children with spina bifida. *Eur J Paediatr Neurol*, 14(1), 86-92.
<http://www.ncbi.nlm.nih.gov/pubmed/19237302>.
doi:10.1016/j.ejpn.2009.01.003
- Wallander, J. L., & Varni, J. W. (1998). Effects of pediatric chronic physical disorders on child and family adjustment. *The Journal of Child Psychology and Psychiatry and Allied Disciplines*, 39(1), 29-46.
- Waterston, T., & Nasser, D. (2017). Access to healthcare for children in Palestine. *BMJ Paediatrics Open*, 1(1), e000115.

- WHO, & Unicef. (2012). Early childhood development and disability: A discussion paper.
- Wilson, R. D., Audibert, F., Brock, J.-A., Carroll, J., Cartier, L., Gagnon, A., . . . Genetics, C. (2015). Pre-conception Folic Acid and Multivitamin Supplementation for the Primary and Secondary Prevention of Neural Tube Defects and Other Folic Acid-Sensitive Congenital Anomalies. *Journal of obstetrics and gynaecology Canada : JOGC = Journal d'obstétrique et gynécologie du Canada : JOGC*, 37(6), 534. [https://doi.org/10.1016/S1701-2163\(15\)30230-9](https://doi.org/10.1016/S1701-2163(15)30230-9).
- World Health Organization, W. (2001). *International classification of functioning, disability and health: ICF*: Geneva: World Health Organization
- World Health organization, W. (2011). *World report on disability 2011*: Press:Geneva.http://www.who.int/disabilities/world_report/2011/report.pdf
- World Medical Association (2013). World Medical Association Declaration of Helsinki: ethical principles for medical research involving human subjects. *Jama*, 310(20), 2191. <http://dx.doi.org/10.1001/jama.2013.281053>.
- Youngblood, M. E., Williamson, R., Bell, K. N., Johnson, Q., Kancharla, V., & Oakley, G. P., Jr. (2013). 2012 Update on global prevention of folic acid-preventable spina bifida and anencephaly. *Birth Defects Research Part A: Clinical and Molecular Teratology*, 97(10), 658-663. <http://dx.doi.org/10.1002/bdra.23166>.